

Linköping University Medical Dissertations  
No. 1278

# ***Living with coeliac disease beyond the diagnosis***

***Susanne Roos***

Department of Social and Welfare Studies  
Division of Health, Activity, Care  
Campus Norrköping, Linköping University  
Sweden



**Linköping University**  
FACULTY OF HEALTH SCIENCES

*Norrköping 2011*

©Susanne Roos, 2011

Cover picture/illustration: Michael Holm

The papers are reprinted with the kind permission of the copyright holders:

Paper I, *Digestive and Liver Diseases*, Elsevier

Paper II, *Scandinavian J Gastroenterology*, Informa Healthcare

Paper III, *J Nursing & Healthcare of Chronic illness*, Wiley-Blackwell

Printed in Sweden by LiU-Tryck, Linköping, Sweden, 2011

ISBN: 978-91-7393-019-2

ISSN:0345-0082

*Till min familj!*

*Det är målet som stöper formen men  
det är vägen dit som formar oss*

*Emma 2011*



# CONTENTS

<b>ABSTRACT</b> .....	<b>1</b>
<b>LIST OF PAPERS</b> .....	<b>3</b>
<b>ABBREVIATIONS</b> .....	<b>5</b>
<b>INTRODUCTION</b> .....	<b>7</b>
<b>BACKGROUND</b> .....	<b>9</b>
<b>Definition of coeliac disease</b> .....	<b>9</b>
<b>Diagnosis and pathogenesis</b> .....	<b>9</b>
<b>Epidemiology</b> .....	<b>11</b>
<b>Clinical feature</b> .....	<b>12</b>
<b>Treatment and adherence to the diet</b> .....	<b>13</b>
<b>Living with a chronic illness</b> .....	<b>15</b>
Everyday life .....	15
Transition .....	16
<b>Aspects of health and well-being in CD</b> .....	<b>18</b>
<b>Self-image</b> .....	<b>20</b>
<b>GENERAL AIM</b> .....	<b>23</b>
<b>METHODS</b> .....	<b>24</b>
<b>Approach</b> .....	<b>24</b>
<b>Participants</b> .....	<b>27</b>
<b>Assessments</b> .....	<b>30</b>
Well-being .....	30
Gastrointestinal symptoms .....	32
Self-image.....	33
The background data .....	34
Register data.....	34
Qualitative material .....	35

<b>Data analyses .....</b>	<b>36</b>
Statistical analyses .....	36
Qualitative interviews .....	37
<b>Ethical considerations .....</b>	<b>39</b>
<b>RESULTS .....</b>	<b>40</b>
Health and well-being prerequisites for a good life .....	40
The women's health care seeking behaviour .....	41
Relationship between well-being, GI symptoms, self-image and comorbidity.....	43
Everyday life .....	45
<b>DISCUSSION .....</b>	<b>49</b>
Results .....	49
Methodological considerations .....	53
Clinical implications .....	58
Research implications.....	58
<b>CONCLUSIONS .....</b>	<b>59</b>
<b>SAMMANFATTNING PÅ SVENSKA.....</b>	<b>60</b>
<b>ACKNOWLEDGEMENT .....</b>	<b>62</b>
<b>REFERENCES .....</b>	<b>66</b>
<b>APPENDIX .....</b>	<b>84</b>
<b>PAPER I - IV</b>	

# ABSTRACT

**Introduction:** Studies show that women living in Sweden treated for coeliac disease have lower subjective health than other women. After showing signs of remission, adults who have coeliac disease and follow a gluten-free diet, are expected to handle the treatment without any further planned follow-up by health care.

**Aim:** The overall aim of this thesis was to study aspects of living with coeliac disease in adults in the years beyond the diagnosis.

**Methods:** Quantitative methods were used in Studies I, II and III. A qualitative content analysis was performed in Study IV.

**Results:** The results show that women with coeliac disease have a lower level of well-being than men with coeliac disease. The women who have coeliac disease reported a high rate of gastrointestinal symptoms, although they followed a gluten-free diet, and they visited health care services more frequently than women who did not have coeliac disease. A low rate of gastrointestinal symptoms, a positive self-image and few comorbidity emerged as factors that positively affected well-being. Worries also seemed to be a companion of women diagnosed with coeliac disease in adulthood, typically evident when socializing with others.

**Conclusion:** This thesis may provide evidence questioning the validity of declaring all women with coeliac disease showing a normalized intestinal mucosa to be in remission, and thus leaving them to self-management.

**Clinical implications:** Health care professionals need to be aware of that the transition to a gluten-free life may vary for individuals. It does not seem enough to

follow a gluten-free diet to reach a state of good well-being for all women. A major task for health care providers is therefore to support women with CD in reaching better subjective treatment outcomes. The results may also contribute to that health care system develops routines in order to optimise the care and treatment of these women.

**Keywords:** Chronic illness, Coeliac disease, Comorbidity, Gastrointestinal complaints, Health care use, Self-image, Well-being.

# LIST OF PAPERS

This thesis is based on the following papers, which will be referred to in the text by their roman numerals.

- I. Roos S, Kärner A, Hallert C. (2006) Psychological well-being of adult coeliac patients treated for 10 years. *Digestive and Liver Disease* 38, 177–182.
- II. Roos S, Wilhelmsson S, Hallert C. (2011) Swedish women with coeliac disease in remission use more health care services than other women: a controlled study. *Scandinavian Journal of Gastroenterology* 46, 13–19.
- III. Roos S, Wilhelmsson S, Vulcan A, Sjöberg K, Hallert C. (2011) Bowel symptoms, self-image and comorbidity impact on well-being of women with coeliac disease. *Journal of Nursing and Healthcare of Chronic illness* 3, 302–309.
- IV. Roos S, Hellström I, Hallert C, Wilhelmsson S. The worried well- Women living with coeliac disease in Sweden. (Submitted).



# ABBREVIATIONS

AGA	Antigliadin antibodies
CD	Coeliac disease
CDWÖ	The Care Data Warehouse of Östergötland County Council
EMA	Endomysial antibodies
GFD	Gluten-free diet
GI	Gastrointestinal
GSRS	Gastrointestinal Symptom Rating Scale
HLA	Human leucocyte antigen
IBS	Irritable Bowel Syndrome
ICD	International Classification of Diseases
ICN	International Council of Nurses
PGWB	Psychological General Well-Being Index
SASB	Structural Analysis of Social Behaviour
t-TGA	Tissue transglutaminase antibodies



# INTRODUCTION

Living with a chronic illness affects a person's entire life and involves physical, psychological and social consequences requiring adjustment in several different life domains (Stanton *et al.* 2007). Coeliac disease (CD) is a lifelong but not life-threatening illness of the gastrointestinal (GI) tract, and affects genetically predisposed gluten-intolerant individuals of all ages (National Institutes of Health 2004). The only treatment known is a gluten-free diet (GFD) for life. CD ranks as a prevalent condition in most Western populations and represents a health issue (Mäki *et al.* 2003, Green & Cellier 2007, Hopper *et al.* 2007, World Gastroenterology Organisation 2007, National Institute for Health and Clinical Excellence 2009, Mustalahti *et al.* 2010). Studies (Ciacci *et al.* 2003, Whitaker *et al.* 2009) show that living with CD and remaining well is far from easy for many people diagnosed in adulthood. Learning to live with a lifelong illness includes a changed self-image (Kralik 2002) and a negative impact on well-being (Lohiniemi 2001). Well-being is a multi-shift concept that is influenced by many different factors, and having a disease is one of them (Ryan & Deci 2001). Swedish women living with CD show a lower degree of well-being than women in the general population and than men with CD, and they report a high rate of GI symptoms for unclear reasons despite following a GFD for years (Hallert *et al.* 1998, Midhagen & Hallert 2003). There is also evidence that suggests that women with CD take more responsibility for planning, shopping for and preparing food than men with CD (Sverker *et al.* 2009). Although CD can be

brought into histological and clinical remission by a GFD, the disorder cannot be cured (Lohiniemi 2001).

The full range of long-term subjective outcomes of adults with CD living on a GFD has not been fully studied. The International Council of Nurses (International Council of Nurses) identified health and well-being as an important topic of interest within nursing research. Studies of health-focused care include, among other things, health prevention, experience of lifelong ill-health, quality of life, and maintaining and safeguarding a person's views about well-being and health (International Council of Nurses 2001).

CD seems to affect the person's entire life, which may reduce health, well-being and self-image, and can be associated with virtually any symptom before the diagnosis is made, which can remain even with the dietary treatment. In this thesis, aspects of living with CD in adulthood are studied, focusing on the impact of the illness on well-being, GI symptoms, comorbidity, and self-image, mainly in women. To provide good quality health care for individuals with CD it is important to study different aspects of what it is like to live with a chronic illness.

# BACKGROUND

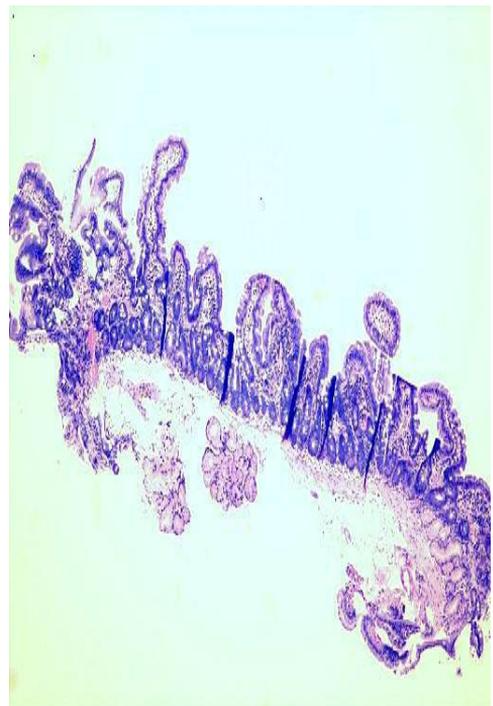
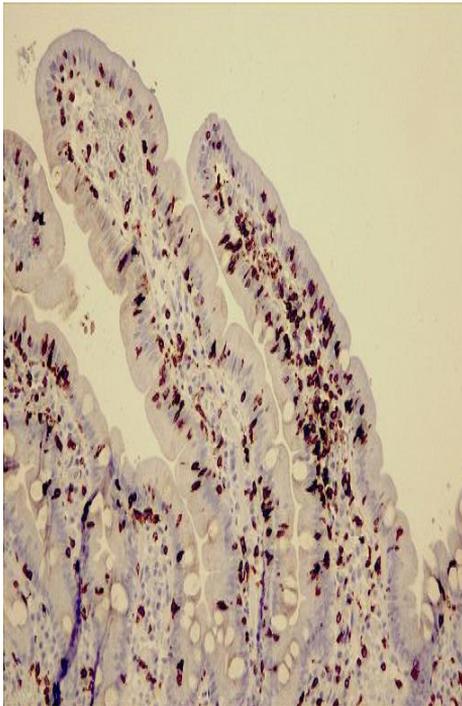
## Definition of coeliac disease

CD can be defined as an autoimmune disorder triggered by an inappropriate response to dietary gluten peptides in genetically predisposed children and adults, resulting in damage to the small intestine, with villous atrophy and crypt hyperplasia as typical features (Mäki & Collin 1997, Ciclitira *et al.* 2001, National Institute for Health and Clinical Excellence 2009).

## Diagnosis and pathogenesis

The diagnosis of CD is based on examination of specimens of the duodenum obtained in the untreated state showing flat or nearly flat mucosa or an increased number of intraepithelial lymphocytes, or both (Abdulkarim & Murray 2003). The diagnosis is strongly supported by positive CD serology (Dipper *et al.* 2009). The diagnosis can be reliably excluded by means of genetic testing showing the absence of HLA haplotypes (Michalski *et al.* 1996). According to an international expert panel in 2007, intestinal biopsies together with a positive serology, preferably anti-tissue transglutaminase antibodies (t-TGA), represent the gold standard for diagnosing CD (World Gastroenterology Organisation 2007). There has been a major development in serological tests and currently antigliadin (AGA) and antiendomysium (EMA) are replaced by t-TGA as the standard test (Grodzinsky *et al.* 1990, 1994, Laurin *et al.* 2004). Prior to the introduction of serological tests, CD was suspected in patients

showing signs of malabsorption that called for an intestinal biopsy to establish the diagnosis. It took several years after the work of Dicke (1950) in the Netherlands for CD to be frequently recognized in Sweden, mainly as a childhood disorder. (Berg & Lindberg 1979, Stenhammar 1981). Subsequently Hallert *et al.* (1983) reported a similar prevalence in Sweden of CD in adults as in children. Today adults suspected to have CD are referred for endoscopy either to confirm or dismiss the tentative diagnosis. The diagnosis of CD is firmly established after clinical and histological improvement following the start of a GFD. Adults diagnosed with CD are strongly advised to continue on a strict GFD for the rest of their lives by self-management, and are recommended to seek primary care services for follow-up as needed. A history of close adherence to a GFD is shown to indicate a normalized intestinal mucosa (Grodzinsky *et al.* 2011).



**Figure 1.** Normal villi (left picture) and villous atrophy (right picture) in the small bowel. (Hallert C, Östergötland County Council)

The current concept of CD postulates that it develops exclusively in individuals with alleles that encode for HLA-DQ2 or -DQ8 proteins. It is estimated that every third person in Western populations carries these alleles but only a few carrying DQ2 develop CD. The HLA restriction in CD is related to the physical properties of gluten. The HLA-DQ2/-DQ8 molecules are responsible for presenting gluten, thereby activating gluten-specific T cells. The glutamine part of gluten is the target for the action of the enzyme t-TGA. As a result, the gluten protein is modified and effectively bound to DQ2 to enhance T-cell recognition (Freitag *et al.* 2004). This concept may be insufficient to explain why genetically predisposed people do not consistently develop CD at first exposure to dietary gluten but present later in life. It may suggest a set of as yet unknown factors accounting for, for example, environmental exposure in the incidence of CD (Ivarsson 2001).

## **Epidemiology**

CD was long classified as a major malabsorption syndrome until the discovery by Grodzinsky *et al.* (1992) of a surprisingly high prevalence (1:256) in apparently healthy Swedish blood donors, thereby drawing attention to clinically silent forms of CD. This led to numerous screening studies including adults as well as children worldwide. With the increased awareness and the introduction of specific sensitive and specific serology tests, CD has been found to be one of the commonest hereditary human disorders. CD, like many other autoimmune diseases, is more common among women (Jacobson *et al.* 1997, World Gastroenterology Organisation 2007) with a female to male ratio of between 2:1 and 3:1, and also seen in children (Ivarsson *et al.* 2003). The prevalence of CD may vary from 0.5 % to 2% in general populations in Europe and the United States (Hopper *et al.* 2007, Virta *et al.* 2009,

Kaukinen *et al.* 2010, Tack *et al.* 2010). A recent study in Sweden, however, revealed that 3% of 12-year-old children in Sweden have CD, probably triggered by rapid and early introduction of gluten (Myléus *et al.* 2009). However, there may be other factors than feeding habits that determine incidence rates (Ivarsson 2001, Laurin *et al.* 2004, Troncone & Auricchio 2007). The rate of CD has been underestimated for many years and the incidence of CD is apparently increasing worldwide (World Gastroenterology Organisation 2007, Walker & Murray 2011).

## *Remission*

The short-term goal of treatment of adults with CD is recovery from gluten-induced features such as weight loss, intestinal damage, nutritional deficiencies, unhappiness and tiredness, often referred to as clinical remission. Removal of gluten from the diet leads to full clinical and histological remission of the intestine as outlined by the paediatrician Walker-Smith (1988).

## **Clinical feature**

There are no typical signs or symptoms of CD although the dermatitis herpetiformis is highly suggestive of concomitant CD (Corazza & Gasbarrini 1995, Woodward 2011). Instead most adults presenting for the diagnosis in clinical practice show nonspecific complaints such as malaise and tiredness or symptoms that may mimic any medical disorders, whereas adults experiencing intolerance to cereals are only rarely found to be true coeliac (Kaukinen *et al.* 2000).

## *Symptoms and signs at presentation for the diagnosis*

Indigestion of the gluten protein causes damage to the small bowel mucosa in CD and may also lead to diverse symptoms and illnesses outside the GI tract (Elfström 2009). Hopper *et al* (2007) and National Institute for Health and Clinical Excellence (2009) list the most common symptoms seen in adults with CD presenting for diagnosis:

**GI presenting symptoms and signs:** Abdominal pain, diarrhoea, steatorrhoea, bloating, nonspecific GI symptoms;

**Non-GI symptoms:** Weight loss, fatigue, anaemia, nutritional deficiencies, osteoporosis, arthralgia, arthritis and myalgia, malignancy, alopecia, amenorrhoea/ recurrent abortion, skin rash (dermatitis herpetiformis) and aphthous ulcers, depression or neurological symptoms. Other disorders, such as type 1 diabetes and other autoimmune disorders can coexist with CD. Once gluten is withdrawn from the diet, the villi return to normal and the malabsorption signs will disappear.

## **Treatment and adherence to the diet**

The only treatment known to be effective in CD is permanent withdrawal of dietary gluten (World Gastroenterology Organisation 2007, Ludvigsson & Green 2011, Woodward 2011). Subsequent studies in Finland and later in Sweden revisiting Dicke's observations concluded that oats can be safely included in a GFD (Janatuinen *et al.* 1995, Störsrud *et al.* 2003). However, people with CD who include large amounts of oats in their diet may experience abdominal pain and discomfort owing to its high fibre content (Störsrud *et al.* 2003, Peräaho *et al.* 2004).

Introduction of a GFD implies profound changes in dietary habits. This can be hard to accomplish for any adult living in countries where gluten-containing foods represent a major part of the traditional dishes. Accordingly, the rates of adherence to the GFD differ widely throughout Europe (Hall *et al.* 2009); in Italy, around 70% of people with CD adhere to the diet while in Nordic populations this figure is close to 90% (Hallert *et al.* 1998, Ciacci *et al.* 2003, Hall *et al.* 2009).

Studies have provided evidence of a safe threshold of daily gluten intake, indicating that ingesting trace amounts (10–20 mg) of gluten is harmless in practice; for comparison, a Western daily diet contains 10–20 g of gluten (Collin *et al.* 2004, Kaukinen *et al.* 2010). This may put into question current routines advising adults to keep to a strict diet, all at risk of imposing fear of gluten contamination in virtually any food (Sverker *et al.* 2005). However, as pointed out by Akobeng and Thomas (2008) and Wild *et al.* (2010), nutritional therapy in people with CD traditionally centres around lifelong withdrawal of all gluten, with less attention paid to the nutritional quality of the diet. There is evidence to suggest that withdrawal of gluten from the diet may result in a nutritionally unbalanced diet (Thompson *et al.* 2005). In Sweden, women with CD have a significantly higher level of intake of desserts, marmalades and candies than women in general, and a low daily intake of dietary fibre and the B vitamins, folates and vitamin B<sub>6</sub> (Grehn *et al.* 2001). The clinical implications of this were demonstrated by Hallert *et al.* (2009) who showed that adults with longstanding CD who took extra B vitamins for six months showed significant improvement in well-being, including reduced anxiety and depression.

# Living with a chronic illness

The term 'chronic illness' is used to describe a lifelong state which is complex and affects a person's daily life (Miller 1992, Lubkin 1995), and it is a human experience of living with illness (Toombs 1992). According to Sachs (1990) it represents the individual experience of discomfort.

## Everyday life

Living with a chronic illness is likely to affect a person's entire life, and has physical, psychological and social consequences, involving several different life domains (Stanton *et al.* 2007). The diagnosis of CD has two immediate consequences for the individual: one is learning to live with the chronic illness and the other is the necessity of a lifelong GFD, both of which involve big changes (Ciacci 2010). According to Charmaz (1991), learning to live with a chronic illness also takes time and the person needs to overcome judgement, questions and feelings of diminished worth. It is also a challenge integrating a chronic illness into the life context along with enjoying a meaningful life (Whittemore & Dixon 2008). A chronic illness has the effect of setting people apart from others and can take over their life. Living with a chronic illness implies more than learning to live with it:

*“struggling to maintain control over the defining images of self and over one's life. This struggle is grounded in concrete experiences of managing daily life, grappling with illness, and making sense of it”* (Charmaz 1991, p. 5).

From experiences of daily life and as a result of trial and error, people also learn about their response to the illness (Kralik *et al.* 2004). People with a chronic illness need to gain control over their situation and over parts of their treatment (Lindsay 1997). Several studies have shown that living with CD and following a GFD affects a person's lifestyle and quality of life in many ways (Lohiniemi *et al.* 1998, Green *et al.* 2001, Ciacci *et al.* 2003, Gregory 2005). Sverker *et al.* (2005) found that living with CD can absorb a person's entire life situation so that the individual is unable to forget about the illness even for a little while. Feelings of being different from the ordinary population and embarrassment when sharing a table with others can lead to anxiety that can affect the life situation for people with CD (Ciacci *et al.* 2003). The illness must be dealt with in everyday life, and it is primarily in the home that it should be handled and lifestyle changes and practical problems solved. Living with a chronic illness sets demands on a person to learn self-management to resolve the disease-specific issues (Kralik *et al.* 2004). The diagnosis of CD may affect the individual's life several years beyond the diagnosis, for example in reduced enjoyment of food, less social activity, frustration over dietary restrictions, and anxiety about their health, as seen in the UK (Whitaker *et al.* 2009). Fera *et al.* (2003) puts forward that signs of anxiety and poor well-being are not to be ascribed to CD itself but to difficulties in adjusting to the life-long nature of the disorder.

## **Transition**

Learning to live with a chronic illness initiates a process that also leads to a change in self-image (Kralik *et al.* 2004). This transitional process can be described as a quest for ordinariness so that illness becomes a part of life (Kralik 2002). Living with chronic illness leads to involvement in an ongoing process of transition towards

incorporating the illness into life (Koch & Kralik 2001, Kralik *et al.* 2006). There are no published studies on the transition into a life with CD in adulthood, which is surprising considering the prevalence of CD and its profound impact on everyday life.

Transition can be described as a movement between two different periods, a passage from one life phase, condition, or status to another (Chick & Meleis 1986) and at the end point of transition, it is important that feelings of mastering the situation and a sense of self-esteem and coping appear (Meleis *et al.* 2000). Transition is a multiple concept embracing the elements of process, time span and perception (Chick & Meleis 1986). The transition process may involve different phases and changes in the individual's identity, relationships, abilities and behaviour. Several essential properties of transition experiences have been identified and these include awareness, engagement, change and difference, time span, critical points and events (Meleis *et al.* 2000). It is not certain that the result of a transition is always positive and leads to the growth of the individual. It could instead end with the individual feeling less valuable. The time span to a new beginning is individual and may vary from a short period to several months (Olsson & Ek 2002). Three factors have been identified that affect the transition in a positive direction and make the transition healthy: subjective well-being, mastering the roles, and prosperous relations (Schumacher & Meleis 1994). Health professionals have a role in facilitating incorporation of the illness into everyday life (Kralik *et al.* 2005, Jacobi & MacLeod 2011).

The transition process for a person with CD can have a significant impact on life and can affect thoughts and feelings, which can also have consequences for health and well-being.

# Aspects of health and well-being in CD

Living with CD is far from easy (Lohiniemi 2001) and may negatively affect health and well-being because following a GFD is not enough to bring health and well-being to people living with CD (Usai *et al.* 2007). Defining health may be difficult as health can be viewed from different perspectives.

The biostatic health perspective is disease-oriented. The philosopher Boorse is a proponent of this perspective and he believes that a person has health when body and mind function in accordance with the nature of a typical pattern. Thoughts that if you are not sick, you are healthy dominate here. We can numerically determine the limits of what 'sick' and 'healthy' mean, and apply these values to the individual (Boorse 1977). The health care in the Western societies are largely built around these ideas.

The holistic health perspective on the other hand emphasizes what the individual is experiencing. Lennart Nordenfelt is a proponent of this perspective. He defines health as:

*“A is completely healthy if, and only if, A is in a bodily and mental state which is such that A has an ability to realize all his or her vital goals, given accepted circumstances”* (Nordenfelt 1995, p. 212).

With this definition, Nordenfelt means that health as a phenomenon is dependent on a person's ability in standard conditions to realize minimal happiness. These standard conditions may vary between individuals, cultures, societies and political

systems. By vital goals, he means goals that are necessary and sufficient for a person's minimal happiness (Nordenfelt 1995, 2001). Theories based on a holistic health perspective assume that people are acting beings in relationships with other people, and that health affects a person's ability to act. Nordenfelt's theory is holistic in two aspects: (1) it takes the whole person as its starting point, not the parts or functions of the individual; (2) it takes the individual's goals and his or her environment into consideration.

Since CD seems to affect a person's entire life after the diagnosis (Sverker *et al.* 2005, Whitaker *et al.* 2009) it is not just health that may be affected but also the person's entire well-being.

Most researchers and philosophers perceive well-being as a whole, comprising several parts (Nordenfelt 1994, Downie *et al.* 1996, Bowling 2005). In order to have well-being, Nordenfelt claims that one must be located in a positive mental condition, which is created, influenced or obliterated by combinations of internal conditions, external conditions and activity (Nordenfelt 1994, 2009). Nordenfelt means that quality of life can be described as subjective well-being, and his concept of happiness can be seen as overall well-being (Nordenfelt 2009 and personal conversation, June 15, 2011). He defines happiness as:

*“Sara is happy with her life as a whole if, and only if, Sara wants her life-conditions to be exactly as she finds them to be”* (Nordenfelt 2009, p. 11).

Happiness is connected to a person's wishes and goals, and is a dimension ranging from complete happiness to complete unhappiness. Well-being covers the whole area of people's positive experiences, from sensations to emotions and moods (Nordenfelt 1994, 2000). Happiness is closely linked to a person's wants and goals and can be said to be a function of the relationship between a person's desires and how they perceive their situation (Nordenfelt 1994, 2000). Therefore, living with a chronic illness such as CD, which seems to affect the entire life of the person, can strongly affect their happiness. CD is an illness that is common in women (Hopper *et al.* 2007) and seems to affect the quality of life of women more than men (Hallert *et al.* 1998).

## **Self-image**

Having a chronic illness such as CD may affect a person's psychological status even if it is treated and in medical remission (De Rosa *et al.* 2004). The impact on both self-image and well-being may be particularly bad since a strong self-image is one of the most important components of well-being (Diener *et al.* 1996).

In this thesis, the term 'self-image' (from the Structural Analysis of Social Behavior (SASB) instrument and the theory behind it) is used in the same way as 'self-identity' (from Kralik) and 'self-concept' (from Charmaz).

Having a chronic illness requires reconstruction of a person's self-image by exploring the limitations or boundaries (Kralik *et al.* 2004) that can affect self-image. Self-image can be defined as the way people behave towards themselves and how they handle themselves in relations with others. Self-image is central to life and is shaped in the

very early years (Stern 1991) and continues to evolve and change throughout life. Experiencing chronic illness may result in self-image becoming tied to the past, present or future (Charmaz 1991). Self-image is not just about how an individual evaluates or describes herself; it is also about how to handle herself. The self is always in development and is never a static final product. By not defining their illness, people try to separate it from their lives and to detach it from their self-image (Charmaz 1991). The way we look at ourselves, and think about ourselves and our bodies is an important part of life. CD seems to be a disorder that absorbs a person's whole life, making it hard to forget the disease even for a moment, and with it also follows the feeling of being different from others (Ciacci *et al.* 2003, Sverker *et al.* 2005). A positive self-image is thought to affect how people take care of themselves. Self-image is included in interpersonal theory and, according to this theory it is formed by the impact of interaction, perceptions and interpretations between individuals (Sullivan 1953). Having a chronic illness may also alter the self-image, which can lead either to development or diminishment of the person (Charmaz 1999). The struggle for control is a struggle for balance in life and, between controlling the illness and becoming controlled by it (Charmaz 1991). A discrepancy between a person's self-image and their social role can be a great burden if the self-image is negative (Reich *et al.* 2008).



# GENERAL AIM

The overall aim of this thesis was to study aspects of living with coeliac disease in adults in years beyond the diagnosis.

## **Specific aims**

To evaluate the psychological well-being using the PGWB index in middle-aged adults with longstanding coeliac disease in proven remission, using a population sample of same age as controls.

To examine if women with coeliac disease living on a gluten-free diet for years perceived poor outcome of dietary treatment results in comorbidity that make them use health care services more than other women.

To examine if gastrointestinal symptoms, self-image and comorbidity are related to well-being in women with coeliac disease and whether a history of coeliac disease in childhood impacts on well-being in adulthood.

To explore how women with coeliac disease experience everyday life and their outlook on their future.

# METHODS

## Approach

In this thesis, a quantitative (I, II, III) and a qualitative approach (IV) were used to obtain a richer and deeper description and understanding of what it is like to live with a chronic illness such as CD. This involves two approaches from different paradigms: the positivistic and the naturalistic paradigms. The positivistic paradigm focuses on the objective and quantifiable and uses deductive quantitative methodology. The naturalistic paradigm focuses on the subjective, the non-quantifiable and what is unique for the individual and uses inductive qualitative methodology (Polit & Beck 2008).

The quantitative approach in studies I and III was applied to describe and obtain knowledge about well-being and factors that may affect well-being and daily life in a large group of participants. For this, data were collected in southern Sweden and a cross-sectional design was used. Study II is a cross-sectional study with data from a register to examine health care use during a period of three consecutive years. All known women with CD in remission diagnosed at two gastroenterology departments were included, using a local reference group. In study IV, a qualitative approach, by means of conventional content analysis, was chosen to explore the experience of daily life and thoughts about the future in women with CD. This method was used to gain an inner perspective of the everyday life of women treated for CD.

For an overview of the design, the number of participants and their age, data collection and data analysis in this thesis, see Table 1.

**Table 1.** Description of the four studies included in the thesis.

	<b>Study I</b>		<b>Study III</b>		<b>Study IV</b>
Design	Cross - sectional	Cross - sectional	Cross - sectional	Cross - sectional	Description
Number of participants	30 women and 21 men with coeliac disease. 104 women and 78 men without coeliac disease.	137 women with coeliac disease. 411 women without coeliac disease.	162 women with coeliac disease.	16 women with coeliac disease.	16 women with coeliac disease.
Age	45 – 64 years	20 – 80 years	23 – 94 years	23 - 72 years	
Data collection	Questionnaire	Register	Questionnaires	Interviews	
Data analysis	Descriptive statistics Nonparametric statistics.	Descriptive statistics Nonparametric statistics.	Descriptive statistics Nonparametric statistics, Multivariate logistic regression.	Conventional content analysis.	

# Participants

The participants in the four studies were diagnosed at a gastroenterology clinic according to standard procedures. The diagnosis was made by an intestinal biopsy. Then the participants were referred to a dietician for advice on GFD, and a follow-up intestinal biopsy was carried out 8–12 months later. After showing evidence of remission, the patients were transferred to primary care according to local routines. Dietary adherence at the time of the studies was ascertained by the participants' declaration that they were following a GFD (study I, III, IV), supported by a biopsy or serological test when feasible (study I). There have been major developments over time with regard to biopsies and serological tests. Therefore, grading of the damage to the mucosa may differ in some cases (e.g. grading according to Alexander, Marsh or a modification of Marsh) as well the serological tests used (e.g. AGA, EMA, t-TGA), depending on when the participant was diagnosed. However, biopsies and serological tests were treated in the same way by the various laboratories and clinics.

Recruitment of participants in study I was done by gastroenterologists. Fifty-one middle-aged adults (30 women and 21 men), aged 45–64 years, diagnosed at six gastroenterology clinics in southeast Sweden in a four-year period were included. The participants were in biopsy-proven remission, or, in the case of nine participants who were unwilling to undergo a repeat biopsy, the absence of AGA and EMA antibodies was used as evidence of remission. The participants had all been on a GFD since the diagnosis and had been followed up every second year according to local routines. All the participants responded to the PGWB questionnaire. The reference group consisted of a general sample of 182 adults; aged 45–64 years (104 women and 78 men) randomly selected from a list of participants in a local health

survey and matched by age and gender. They were contacted by mail. In study II, the participants were selected from a list by hospital-based dieticians and the inclusion criteria were: women aged 20 years or more with the diagnosis established by a gastroenterologist and having seen a dietician for dietary advice and information about the GFD. They had all been undergone a repeat biopsy after 8–12 months on the diet. The study comprised 137 women aged 20–80 years with biopsy-proven CD diagnosed at two gastroenterology departments in southeast Sweden. The women had been diagnosed during an eight-year period (January 1995 to December 2002) and were assumed to have been treated in accordance with local routines. Data were collected a minimum of 12 months after diagnosis (from January 2004 to December 2006) to ensure enough time (i.e. at least 12 months) to prove remission and that treatment had lasted for at least one year because it takes around a year for the intestinal mucosa to heal on a GFD. The reference group comprised 411 women living in the same county who had had at least one visit to health care services provided by Östergötland County Council during 2004–2006. For each woman with CD, three controls matched for age and place of residence were randomly selected, carefully excluding women with ICD code K90.0 (coeliac disease). In study III, the participants were recruited by gastroenterologists and a hospital-based dietician. The inclusion criteria were: women aged 20 years or more with biopsy-proven CD, had CD for at least five years, and able to speak Swedish fluently. The study comprised 162 women with biopsy-proven CD diagnosed at a gastroenterology department in southern Sweden. All women were treated according to the local routines for CD. Study IV included 16 women aged 23–72 years living in southeast Sweden with the following inclusion criteria: minimum age 20 years, diagnosed with CD by biopsy at a

gastroenterology clinic, following a GFD for at least two years and able to speak Swedish fluently. The women were recruited from a local coeliac society.

The sociodemographic variables of women and men with CD and the reference groups in studies I and II are shown in Table 2.

**Table 2.** Sociodemographic data of the participants in the four studies included in the thesis.

Variables	Study I		Study II	Study III	Study IV
	Women	Men			
<b>Age groups</b>					
20-39	-	-	32	49	4
40-59	25	17	37	51	5
60-79	5	4	56	56	7
80-99	-	-	12	6	-
<b>Occupational status</b>					
Full-time	16	17	-	-	-
Part-time	11	4	-	-	-
Retired	3	0	-	-	-
<b>Marital status</b>					
Cohabitation	25	16	-	117	10
Single	5	4	-	45	6
<b>Education</b>					
Elementary or occupational school	-	-	-	45	3
Upper secondary school	-	-	-	63	3
University	-	-	-	54	10
<b>Matched reference group</b>	104	78	411	-	-

# Assessments

The different data collection methods used in the studies are described in Table 3.

**Table 3.** An overview of how data were collected in the four different studies.

Assessments	Study I	Study II	Study III	Study IV
Psychological General Well-being index (PGWB)	X		X	
Gastrointestinal Symptom Rating Scale (GSRS)			X	
Structural Analysis of Social Behaviour (SASB)			X	
Background data form			X	
Register data from The Care Data Warehouse (CDWO)		X		
Interviews				X

Details of each collection method appear below.

## Well-being

Well-being was measured using the generic Psychological General Well-Being (PGWB) questionnaire which is designed to assess subjective well-being, distress and the inner personal state. PGWB was selected for this study because it is well validated and has undergone reliability testing on people with upper GI diseases, not only in Sweden but also throughout Europe and the United States.

The scale was first designed for use in a large study in the United States with the aim of providing an index that measures self-reports of intra-personal, emotional or affective states that could reflect a sense of subjective well-being (Dupuy 1984, Bowling 2005). The scale can be used as a one-dimensional outcome indicator using

the total score, although the scale also includes symptoms of disability and of quality of life in different items (Bech 1993). The PGWB consists of 22 items, each using a six-point scale from 1 to 6. The PGWB index is the total score of the six subscales: Anxiety (five items), e.g. nervousness, tension, anxiety, relaxedness and stress; Depressed mood (three items), e.g. depressed, downhearted and sad; Positive well-being (four items), e.g. general spirits, happy, interested in daily life and cheerful; Self control (three items), e.g. firm control, afraid of losing control, emotionally stable, sure of self; General health (three items), e.g. bothered by illness, healthy enough to do things, concerned about health; and Vitality (four items), e.g. energy, awakes rested, feeling fresh, feels active, versus dull, feels tired, worn out. These six subscales provide an index ranging from 22 (the worst possible level of well-being) to 132 (the best possible level of well-being) (Glise *et al.* 1995, Bowling 2005, McDowell 2010). Values  $\geq 100$  indicate a normal or high level of well-being (Dimenäs *et al.* 1996).

The questionnaire has good internal consistency and homogeneity in terms of what it measures. The internal consistency for the six subscales of PGWB as assessed by Cronbach's alpha is: Anxiety 0.82; Depression 0.89; Well-being 0.88; Self-control 0.76; General health 0.61; Vitality 0.85; and for the whole questionnaire 0.90. The instrument has been shown to have high construct validity (Dupuy 1984, Naughton & Wiklund 1993, Bowling 2005) and good face validity since the participants could easily rate their perception. The major strengths of the instrument are that it is a general measure of well-being, and hence is not condition-specific. The instrument also has questions consistent with Nordenfelt's theories on health, such as question 6, 'Did you feel energetic, alert and vital in the past week?' and question 13, 'Have you been worried or concerned about your health during the last week?' Other

questions related to Nordenfelt's theory of well-being, such as question 9, 'Have you felt happy, satisfied and happy with life in the past week?' and question 20, 'I have felt happy and light-hearted during the last week?' (personal conversation, Nordenfelt June 15. 2011).

## **Gastrointestinal symptoms**

GI symptoms were assessed using the Gastrointestinal Symptom Rating Scale (GSRS). The instrument contains five separate GI scales and each scale is composed of two to four highly intercorrelated gastrointestinal syndromes (Dimenäs *et al.* 1995, Wiklund 1995). The questionnaire consists of 15 items combined into the five subscales: Abdominal pain (three items), Reflux (two items), Indigestion (four items), Diarrhoea (three items) and Constipation (three items).

The GSRS is a validated and reliability tested self-administered questionnaire frequently used in GI research and often together with the PGWB index (Dimenäs *et al.* 1995, Dimenäs *et al.* 1996, Wiklund *et al.* 2006). The internal consistency for the dimensions measured with Cronbach's alpha is: Abdominal pain 0.60, Reflux 0.72, Indigestion 0.79, Diarrhoea 0.83, Constipation 0.84, for the dimensions and 0.85 for GSRS in total.

The responses were graded on a seven-point scale. The highest score, 7, denotes the most pronounced symptoms and 1 denotes no symptoms. The higher the score the more pronounced was the symptom. A value of >1.6 indicates a high rate of GI symptoms (Dimenäs *et al.* 1996).

## Self-image

Self-image was determined by the Structural Analysis of Social Behavior (SASB). The SASB model was developed by Lorna Smith Benjamin in the United States in 1996 (Benjamin 1996). The model is based on interpersonal theory, focusing in particular on examining how individuals interact with their environment. Swedish researchers then developed a Swedish version of the SASB model together with Benjamin (Armelius 2001). The questionnaire is designed to give an objective measure of people's self-image and relationships with others. The SASB model has been used in a wide variety of contexts (Benjamin *et al.* 2006). The SASB consists of 36 questions divided into eight different clusters to depict how sympathetic a person is to herself. The responses are given on a scale from 0 to 100, where zero represents "not true at all" and 100 represents "completely true." Cluster 1 relates to autonomy (five items), 2 to self-acceptance (four items), 3 to self-love (five items) and 4 relates to self-nourishment (four items), 5 signals control and self-monitoring (five items), 6 connects to self-neglect (four items), 7 connects to self-rejection (five items) and 8 to self-indictment (four items). The SASB shows whether the person's self-image is reported as positive (high score on clusters 2–4) or negative (high score on clusters 6–8) and whether the self-image is characterized by spontaneity (cluster 1) or self-control (cluster 5). High scores on clusters 2, 3, 4 and low scores on clusters 6, 7, 8 and middle scores on clusters 1 and 5 indicate a positive self-image (Benjamin 1996, Armelius 2001, Meurle-Hallberg & Armelius 2006). For the Swedish cluster model of SASB; reliability is calculated for the cluster version,  $r = 0.87$  (Armelius 2001) calculated using the split-half method and Spearman-Brown correction.

## **The background data**

The background data in study I was collected at enrolment of participants in the study by the local gastroenterologist. For study III, a background data form was sent out to the participants (see appendix). The background data form covered age, marital status, education, occupation, state of health, history of CD diagnosis and symptoms, adherence to the GFD and scheduled follow-up. A pilot test of the form was done using two persons, and none of the questions were changed as the respondents found the questions easy to answer. The background data in study IV were collected during the interview by the interviewer.

## **Register data**

Register data were derived from the CDWÖ, a population-based, diagnosis-related administrative database containing routine health care data such as diagnoses, types and dates of outpatient and inpatient care, and primary health care services run by Östergötland County Council. Medical diagnoses are recorded according to the International Classification of Diseases (ICD-10), and the main diagnosis in addition to secondary diagnoses may be recorded for each patient. Data are transferred to the database monthly.

## Qualitative material

Content analysis (Krippendorff 2004) was used to analyze the interviews. Qualitative content analysis was done in steps in order to find meaning and understanding in the text and the words that have been collected (Krippendorff 2004, Hsieh & Shannon 2005).

The participants were told to speak freely on the following topics: What is it like to live with CD? This initial question was put to all interviewees, then, depending on how the interviews developed, the following two questions were asked: Does the disease affect your daily life, how/why? How do you look upon the future? All questions were followed up by supporting questions such as: Can you tell me more/develop? Do you have something to add? Can you give examples? How do you mean now?

Demographic questions were asked at the end of the interview. These covered age, occupation, the year of the CD diagnosis, who made the diagnosis, marital status and heredity, and also who the participants talked to when facing problems with the disease.

The interviews lasted between 26 and 65 minutes; 15 lasted 30 minutes or more. The interviews were all conducted by this author. The interviews were performed in the women's homes ( $n=5$ ) or in a neutral location ( $n=11$ ) according to the woman's preference. All interviews were carefully transcribed word by word as a whole, including hesitations, pauses, laughter and stronger emphasis of words. The interviews were transcribed by this author ( $n=10$ ) and one medical secretary ( $n=6$ ).

# Data analyses

## Statistical analyses

Nonparametric methods were used since the data from Studies I–III were not normally distributed. Statisticians assisted with the statistical analyses. Data were presented as means and 95% confidence intervals (CI) (study I,III) in order to enable comparisons with the results of previous studies using PGWB and GSRS and median and SD (study II). Demographic data were reported as absolute and relative frequencies (studies I, II, III). A nonparametric method, the Mann-Whitney *U* test, was used to compare two independent groups or subgroups in studies I, II, III, and in study III the Kruskal–Wallis test was used for comparison of three independent groups. The chi-squared test was used to analyse proportions. The use of health care services (number of visits) in study II was estimated by calculating the number of annual visits by the women with CD and female controls during 2004–2006 to any health care service run by Östergötland County Council. The calculation was carried out by dividing the total number of patient visits by the number of exposed days (i.e. days living in Östergötland County) during those years. The total was multiplied by 365 days. The data analysis included all ICD-10 categories of disease, excluding categories containing mainly infectious diseases, injuries and nonspecific diseases. Using multivariate logistic regression in study III, the PGWB index was dichotomized into high/normal well-being or low well-being, and used as a dependent variable. The odds ratio was calculated for GI symptoms (low rate versus high rate), self-image (positive versus negative), comorbidity (no disease versus one or more diseases besides CD) and onset of CD symptoms (in childhood versus in adulthood). All calculations were done by means of SPSS® for Windows version 14, 17 and 18

(Statistical Package for the Social Sciences, SPSS Inc., Chicago, IL). The significance level was set at  $p < 0.05$  (double-sided).

## Qualitative interviews

A conventional content analysis as described by Hsieh and Shannon (2005) was used for the subjective interpretation of the content.

- All data was first read several times so that the researcher could become immersed in the data. The text was read as a story so the overall feeling of the data emerged.
- The text was read again line by line so that the author could find key words to assist with generating codes in the text, thus allowing the text mass to be reduced.
- The key words from the material were written down in order to capture key thoughts to describe all aspects of the content. This became a preliminary analysis of the collected material.
- Labels were drawn from the text to the codes, and the words of the informants were used for the labels. These labels could consist of several thoughts and keywords and they could reflect more than one key thought. These labels were used to build the first coding scheme.
- These codes were grouped to build categories. The codes were sorted into different categories according to the way they connected to each other and how they were linked together.
- These categories then became clusters, leading to nine subcategories and six categories for synthesizing themes of meaning.
- Finally after discussions, themes were formulated.

In qualitative research, the terms of validity and reliability are called trustworthiness (Credibility, Conformability, Dependability, Transferability) (Patton 2002, Polit & Beck 2008). To ensure the trustworthiness of the data, this author analysed the data and formed subcategories and categories from the interviews and formulated themes for these interviews. The three co-authors then read the transcript interviews to confirm first the keywords and then the categories that were created from the data at several meetings. One author had an extensive pre-understanding of CD, and did not take part in the analytical process (Credibility). The first author of the paper had previous experience in working with science around persons with CD. To balance the pre-understanding, two of the co-authors read the interviews with special care because they did not have the same experience of CD that this author had (Conformability). The dependability was secured by ensuring that all transcriptions were proofread by the interviewer and verified from the tape recording. Transferability was addressed by giving a description of the women, the environment, the interviews and by using quotations so that the reader could form a view on transferability.

## **Ethical considerations**

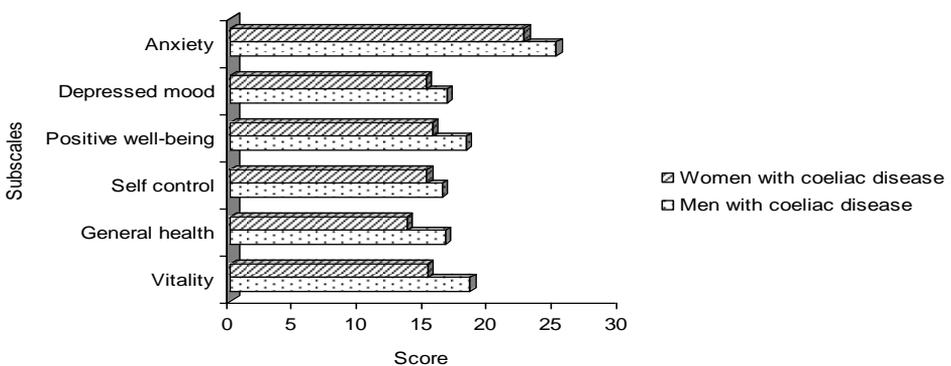
The studies included in this thesis were conducted in accordance with the Declaration of Helsinki (1964) and were approved by the Regional Ethical Review Board Linköping University, Linköping, Sweden: study I, number 96068; study II, number M14-08-T56-09; study III, number M2010/63-32; and study IV, number M188-09. The studies were also approved by the heads of the participating departments. Informed consent was obtained from the participants prior to the studies. There was no dependent relationship between the participants and the researchers that could have influenced any participant's decision to participate in the studies. They were also given written information on all studies and verbal information on study IV. It was made clear that participation was voluntary and that they were free to withdraw from the study at any time and confidentiality was guaranteed. The integrity of the participants was taken into account and they were also assured that it would not be possible to identify them. Conducting research on humans always carries a risk of invading other people's lives and privacy. This could have unseen consequences for the participants, and their interests had to be put before the research (Polit & Beck 2008). The studies included in this thesis were done with this in mind, while still trying to describe the impact that living with CD had on the everyday lives of the participants.

# RESULTS

## Health and well-being prerequisites for a good life

Study I found that women with CD showed no statistically significant difference in well-being either on the different subscales of PGWB or the total PGWB index, 97.1 (95 % CI = 90.6 - 103.7) compared with the female reference group 102 (95% CI = 99–106).

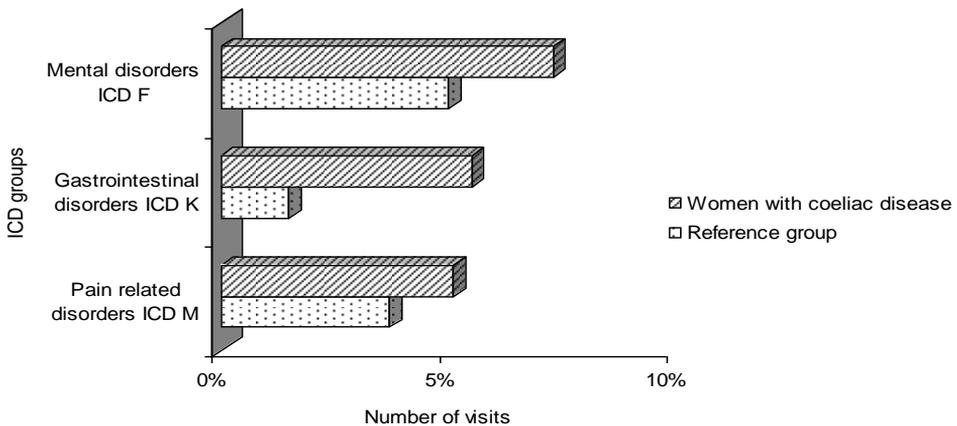
The comparison between men and women with CD showed that men's higher scores on the subscales of the PGWB (Figure 2) resulted in a statistically significant higher total PGWB index score 111.1 (95 % CI = 105.2 - 116.9) vs. 97.1 (95 % CI = 90.6 - 103.7) ( $p < 0.003$ ).



**Figure 2.** Mean score for men and women with CD on the different subscales of the Psychological General Well-Being index ( $p < 0.05$  in all subscales except for self control)

## The women's health care seeking behaviour

The main findings in study II were that women with CD in remission used more health care services in mean 9.1 per year ( $\pm 12.1$  SD) than the female reference group of the same age, mean 7.1 per year ( $\pm 18.7$  SD) ( $p < 0.05$ ). The main conditions prompting women with CD to seek health care were mental and behavioural disorders (ICD F), diseases of the digestive system (ICD K) and diseases of the musculoskeletal system (ICD M) ( $p \leq 0.01$  for all three comparisons), omitting diagnostic groups containing infectious diseases, injuries and nonspecific diseases (Figure 3).



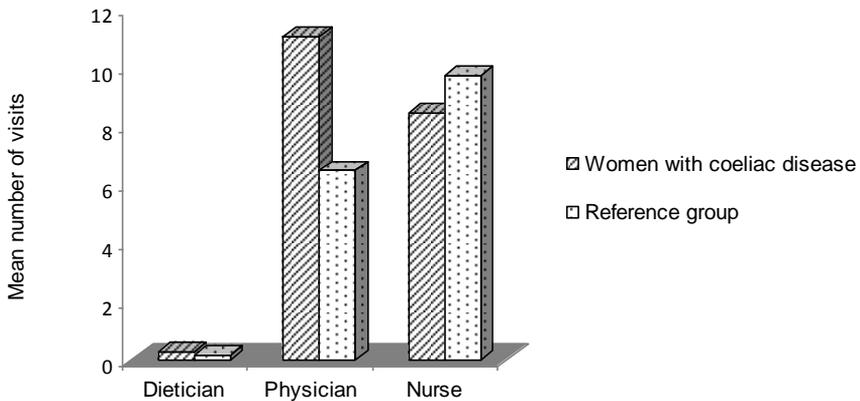
**Figure 3.** The proportions of health care visits of mental, gastrointestinal and pain related disorders in women with CD compared with the female reference group

The women with CD who sought health care for mental disorders were mostly diagnosed with depression, stress disorders and bipolar disorders. For GI disorders, the majority were diagnosed with CD, functional disorders and inflammatory bowel diseases. The women with CD who sought help for pain-related disorders were mostly diagnosed with myalgia, arthralgia or nonspecific arthritis. The reference

group presented most for diseases of the musculoskeletal system, cardiovascular diseases and malignant disorders. No obvious difference in health care use was seen in regard to age.

The number of women visiting health care providers because of GI symptoms was significantly higher for the women with CD than for the female controls (53.7% vs. 15.5%), whereas the number of individuals making several visits to health care providers did not differ between the three ICD groups. The distribution of individual health care use for mental, GI and pain related disorders were similar for women with CD and the reference group.

Figure 4 depicts the mean number of visits to dietitians, physicians and nurses for women with CD and for controls.



**Figure 4.** Mean number of visits to dietitians, physicians and nurses for women with CD and for the female reference group during a period of three years

The higher frequency of visits to health care providers for women with CD mainly involved visits to physicians in primary care.

## Relationship between well-being, GI symptoms, self-image and comorbidity

Study III addressed factors that may influence the well-being of women living with CD and studied relationships between well-being and GI symptoms, self-image, comorbidity and a childhood history of CD. The multivariate regression analysis (Table 4) showed that well-being was negatively related to a high rate of GI symptoms, a negative self-image and comorbidity, whereas a childhood history of CD showed no relation to well-being in adulthood.

**Table 4.** Odds ratios (ORs) for well-being of women with CD with high ( $n = 77$ ) versus low levels ( $n = 85$ ) of well-being: results from multivariate logistic regression analyses.

Dependent variable	Independent variables	Value	Multivariate*		
			OR	(95% CI)	p-value
Well-being	Gastrointestinal symptoms	low↔/high	16.9	(4.5-63.1)	0.001
	Self image	positive↔/ negative	6.0	(2.1-16.9)	0.001
	Comorbidity	no↔/yes	2.8	(1.2-6.4)	0.019
	Symptoms since childhood or not	no/yes↔	2.1	(0.9-4.8)	n.s

CI= confidence interval; \* all independent variables in the model; ↔ reference category

Most of the participants were under 60 years of age (Table 1) and the study revealed that 62% of the women with CD stated that they had at least one disease apart from their CD. Forty percent stated their present state of health was bad.

The mean values and CI for PGWB index and reference values (Dimenäs *et al.* 1996) are shown in Table 5.

**Table 5.** Mean values and 95 % Confidence intervals (CI) of well-being in women with CD and reference values.

Subscales	All women n=162		Reference values*	
	Mean	(CI)	Mean	(CI)
Anxiety	22.36	(21.51-23.21)	23.62	(23.31-23.92)
Depressed mood	14.62	(14.13-15.11)	15.31	(15.13-15.49)
Positive well-being	15.12	(14.49-15.76)	15.95	(15.70-16.21)
Self-control	14.24	(13.74-14.74)	15.06	(14.88-15.23)
General health	13.08	(12.51-13.66)	14.41	(14.21-14.61)
Vitality	14.99	(14.26-15.72)	16.95	(16.69-17.21)
Total score	94.45	(91.14-97.75)	101.41	(100.23-102.60)

\*Dimenäs *et al.* 1996

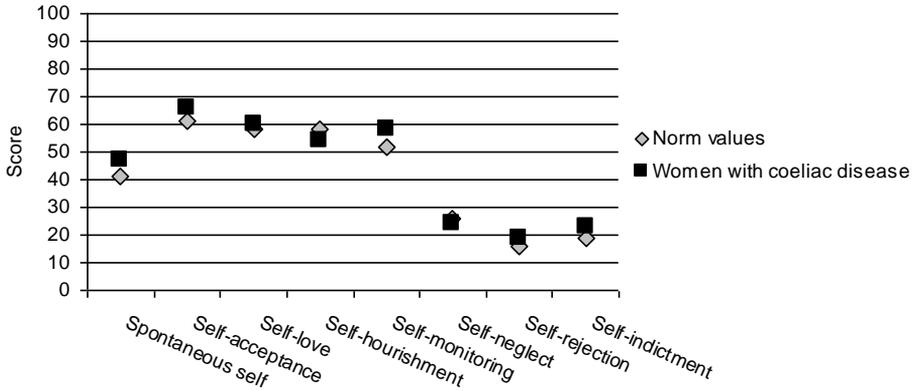
The mean values and CI for GSRS scores and reference values (Dimenäs *et al.* 1996) are shown in Table 6.

**Table 6.** Mean values and 95 % Confidence intervals (CI) for gastrointestinal symptoms in women with CD and reference values.

Subscales	All women n=162		Reference values*	
	Mean	(CI)	Mean	(CI)
Abdominal pain	2.35	(2.17-2.53)	1.63	(1.58-1.68)
Indigestion	2.71	(2.53-2.89)	1.78	(1.73-1.83)
Reflux	1.94	(1.75-2.13)	1.37	(1.33-1.42)
Constipation	2.42	(2.20-2.65)	1.65	(1.59-1.71)
Diarrhoea	2.35	(2.10-2.60)	1.39	(1.35-1.43)
Total score	2.41	(2.26-2.56)	1.56	(1.52-1.59)

\*Dimenäs *et al.* 1996

Figure 5, depicts the mean values of self-image in the entire study population, together with the norm slope for positive self-image (Meurle-Hallberg & Armelius 2006).



**Figure 5.** Mean values of self-image in women with CD compared with the norm slope for the Structural Analysis of Social Behaviour scale

Seventy-two percent of the women with CD had a positive self-image.

## Everyday life

Study IV was designed to explore the everyday life of women living with CD and their outlook for the future. The findings indicate that living with CD is complex and concerns the entire life, ranging from the past to the present, and also includes thoughts about the future. In the content analysis, three themes stood out: Illness trajectory and treatment, Socializing with others, and Feeling of loneliness and worries. The themes, categories and subcategories are shown in Table 7.

**Table 7.** Subcategories, categories and themes from the content analysis.

<b>Subcategories</b>	<b>Categories</b>	<b>Themes</b>
Abdominal discomfort and fatigue Relieved or feeling disappointed	Diagnosis	Illness trajectory and treatment
Being prepared and meticulous	Dietary treatment	
Limits in life	Social limitations	Socializing with others
Bothering Sadness and vulnerability	Alienation	
Being invisible Feeling of loneliness	Feelings	Feeling of loneliness and worries
Worried well	Lack of understanding	

The main finding of this study was that worries are a close companion of women diagnosed with CD in adulthood, and are typically evident when socializing with others.

In the theme, Illness trajectory and treatment, most of the participants perceived that a lot of different symptoms, mainly fatigue and symptoms of the GI tract, had been present throughout their life. The diagnosis of CD brought relief to some because they were reassured that it was nothing worse. For others, the diagnosis soon became a disappointment when they discovered that symptoms tended to persist or even develop after starting the GFD. The women also expressed perceptions of constant suffering when they did not get any better. They also described how they always had to be prepared and carefully stick to dishes without any gluten. They used statements saying, for example, that they would be “*sick every other day if only*

*they would take a chance and buy just anything for food*". The women were extremely careful to scrutinize the ingredients of every food package to be sure there was no gluten in it. This reading took much time but it was worthwhile because accidentally ingesting something containing gluten would make them very sick, with many symptoms. They could all tell the exact time when they last ingested something containing gluten and what had happened. The women who had been diagnosed in childhood expressed fewer concerns about the restrictions associated with eating a GFD; as a consequence they found it easier to manage the diet than the other women. Cooking at home was usually no trouble for any of the women, so they brought lunch boxes from home to work. They were all prepared, with extra food in their bag so they could always have something with them (ranging from fish to special biscuits) and at home if they were unable to cook something suitable. This preparation also applied when they knew they might have to dine out. They resorted to extraordinary planning and even called restaurants to ensure that they served some gluten-free food. All of this set profound social limitations because they were rarely able to do anything spontaneously.

The theme, *Socializing with others*, describes various limitations in living with others. The women became apprehensive when going out, and therefore did not dare to participate in activities with others. Much of their social life became less enjoyable as soon as there was a focus on eating outside the home; eating at home with others was far less troublesome. This theme also encompassed how some participants felt unable to enjoy travelling to various countries because they did not want to risk getting sick by eating local dishes, sticking to salads for a week instead. The theme also covered the limited choice of cakes when they had coffee with friends, which

resulted in just biscuits and ice cream, evoking a sense of grief. Proving and at times bothering that they were different from others added to feelings of sadness and increased vulnerability.

The theme, Feelings of loneliness and worries, encompasses a feeling of not being nurtured by health care. The women lacked security because no one cared after they received their diagnosis and were declared to be in remission. The women all expressed a need for regular follow-ups, not only through blood samples but also by seeing health care professionals with an interest in CD. The decision to withdraw financial support to adults having to follow a GFD was a matter of much anger among the women, reinforcing the sense of being invisible to society. Some also referred to the practice of regular follow-up of people with diabetes. They also expressed worries that following a GFD for life might not be nutritionally satisfactory in every dimension and may even account for ill-health now and in the future. However, most thoughts about the future were related to worries about getting old and losing control of the content of their everyday diet and hence not be taken seriously about their gluten sensitivity. They expressed sorrow that health care professionals failed to show insight into what it really means to not feel well despite carefully adhering to a GFD and how it is to live with a GFD.

# DISCUSSION

## Results

The present thesis focuses mainly on women since they have been found to perceive the outcome of the dietary treatment as worse than men with CD do. This finding was supported when it was shown that the women had a lower PGWB Index than the men, indicating a generally lower level of well-being i.e. consistent with Ciacci's (2003) ideas of fear, anger, anxiety and sadness, accompanying CD in adulthood. This signalled that reduced well-being is a feature of women living with CD for years. This intriguing finding may agree with the observation that women living with CD perceive the overall burden of their condition as distressing in terms of interfering with socializing with friends, having to abstain from important things in life, including fears that the children may develop CD (Hallert *et al.* 2003). In addition to this, for some women, having CD brings feelings of being lonely and different and invisible to others, which affects their everyday life. Moreover, their concerns about the future, and worries about present and future health despite being declared in clinical remission go generally unnoticed by health care providers. This would allow for comparisons with other chronic illnesses using questionnaires designed to explore well-being and disease-related worries and concerns (Stjernman *et al.* 2010).

It is clear that eating a GFD for years is not enough for all women with CD to feel well. Furthermore, living with an illness such as CD seems to affect a person's entire life regarding bodily symptoms, food, frustrations over limitations and fewer choices

(Sverker *et al.* 2005, Whitaker *et al.* 2009). According to Nordenfelt (1994, 2000), well-being covers people's positive experiences, from sensations to emotions and moods. According to Charmaz (1991), living with a chronic illness means more than learning to live with it, it takes time and perseverance to manage everyday life. The transition to a life on a GFD may be a component that is overlooked; life with CD possibly affects women diagnosed in adult life more than previously thought. It is likely that women with CD diagnosed in adult life have difficulty in coping with their illness. Suggesting that the transition to a life with a GFD grows harder with increasing age at diagnosis is consistent with the observation that children diagnosed in the first years of life find it easier to follow the GFD (Högberg *et al.* 2003). However, adults with CD living in Finland do not appear to look upon the dietary treatment as burdensome (Kurppa *et al.* 2010) and generally show improvement in subjective health (Ukkola *et al.* 2011). Yet keeping to a strict treatment regime is well understood to take its toll in the area of well-being, as witnessed in adults struggling with type I diabetes. The more closely one is to strictly adhere to the diet, the harder it seems to incorporate the disease (Karlsson 1997).

The association between CD and mood disorders has been studied frequently (Ciacci *et al.* 2002, Accomando *et al.* 2005, Ludvigsson *et al.* 2007) but remains largely unexplained. Ludvigsson (2007) and Arigo (2011) showed that depression is often associated with CD, not only in the untreated state. It is possible that personality traits, family history, comorbidity and nutritional deficiencies, notably lack of B vitamins, all contribute (Goldberg 1970, Hallert 1981).

Women with CD declared to be in clinical remission are routinely left to manage CD on their own with no regular follow-up. This is based on the belief they will remain well in the course of treatment. Somewhat surprisingly, they appeared to consume health care for mental disorders, GI disorders and musculoskeletal diseases to a greater extent than women of similar age in the general population. This cannot be readily explained by dietary transgressions as studies have reported a very high degree of adherence to the GFD among Swedish women (Hallert *et al.* 2003, Midhagen & Hallert 2003), which has proved to be a fairly good marker of sustained histological remission in CD (Grodzinsky *et al.* 2011). Interestingly it would appear that CD is frequently associated with comorbidity other than disorders known to be genetically linked with CD such as microscopic colitis (Stewart *et al.* 2011), all raising questions about the ability of self-management of women with CD to promote and sustain subjective health. Exploring factors that impact the well-being of women with CD revealed that it is negatively affected by negative self-image, GI symptoms and comorbidity. In particular, the high prevalence of irritable bowel syndrome (IBS) - like symptoms probably accounts for much of the perceived ill-health of patients with CD (O'Leary *et al.* 2002, Häuser *et al.* 2007, Usai *et al.* 2007). Interestingly the health care seeking behaviour of women with CD in remission is similar to that of women with functional GI disorders (Hillilä *et al.* 2007), and female gender is also a predictor of constant worries in CD (Häuser *et al.* 2010).

A recent review clarified that an experienced and properly trained nurse is able to produce as high quality care as primary care doctors concerning the health outcome of the patient (Laurant *et al.* 2009). To date, however, nursing appears to be a neglected component in the follow-up of adults with CD in Europe as well as in the

United States (Bebb *et al.* 2005, Pietzak 2005). However, a recent randomized controlled trial showed promising results with structured nurse-led education of women living with CD (Jacobsson 2011). Overall, the present results would imply a need of launching multi-disciplinary teams, as proposed by Silvester & Rashid (2007).

### *In Summary*

The results of the present studies may be taken to put into question parts of the current management of women living with CD. The management tends to solely focus on eating a strict GFD to keep intestinal mucosa free of inflammation (Parnell & Cicillitra 1999, Abdulkarim & Murray 2003, World Gastroenterology Organisation 2007, Haines *et al.* 2008) instead of taking in to account the worries experience of women with CD. After all, only a strict diet proves insufficient to ensure health and well-being to all women.

## **Methodological considerations**

This thesis concerns an illness that over the years has attracted little research in the field of nursing. The methodological approach was selected to extend our understanding of CD beyond diagnosis by applying an approach within the framework of nursing. Three of the studies are quantitative and the fourth study uses a qualitative content analysis approach.

There are some strengths and limitations of the thesis needed to be taken into considerations.

### *Strengths and limitations*

The inclusion criteria used required a minimum of two and five years on a GFD, suggesting that the time spent on the diet differed widely in the populations under study. This may imply that the improved availability of gluten-free foods over the years has made CD easier to live with for women diagnosed in recent years, although evidence is lacking that this has increased the rate of strict adherence to the diet or the subjective outcomes of treatment. The procedure used in gastroenterological clinics for diagnosing CD in Sweden has not undergone any major changes in the last 20 years

Studying mainly women with CD may limit the conclusions. On the other hand, we were careful to include women from different parts of Sweden in order to minimize selection bias. The decision to exclude men with CD was based on the observation that they appear to find it much easier to live with CD than women. Interviews with

men who have CD might have been appropriate to gain further knowledge in the field.

## *Participants*

The participants were recruited from seven different hospitals in southern Sweden (Studies I-III) and were assumed to have been diagnosed by means of intestinal biopsies and eating a GFD for years, implying they had attained a healed intestinal mucosa and were unlikely to have non-responsive CD.

The great majority of the participants were transferred to primary care for follow - up in accordance with local routines for managing newly diagnosed adults with CD showing signs of remission.

It should be noted that study II was a register based study with no review of the patients' records was made.

Studies I and II included reference groups (larger than 1:3 study I and 1:3 in study II) but power calculations were done only in study I. The fact that the statistical power was not calculated may be a limitation that should be kept in mind when discussing the results.

The conclusions drawn might have been somewhat too general because the results of study I concern just middle-aged men and women, and the size of the study population was limited. The results of studies II and III are believed to be valid for adult women with CD in general, considering the study population ranged in age

between 20 and 94 years. In study IV, the size of the study population would seem small yet the interviews were relatively long and yielded a large quantity of coherent information without facing validity problems. To this end no rules are stated as to the number of participants in qualitative research studies (Patton 2002, Kvale & Brinkman 2009).

## *Design*

An inherent limitation of a cross-sectional design (studies I, II and III) is that it is restricted to studying a phenomenon at one point in time only and the results should therefore be interpreted with caution (Polit & Beck 2008). Well-being in particular is a shifting phenomenon, and may vary greatly over time. However, the health care seeking patterns found provide support for the overall results of the thesis.

Studying living with an illness and well-being was thought to be suitable for a quantitative approach using validated and reliable questionnaires. It could be argued that the results may reflect operational definitions, and a qualitative approach as used in study IV adds a personal view (Berg 2004) of living with CD and perceptions related to well-being.

## *Assessments*

In the present thesis four different questionnaires were used. The value of self-reported data in questionnaires may be limited since some participants may want to give a good impression (Polit & Beck 2008) or may not give every question their full attention if there are many questions (Lind 2001, Ejlertsson 2003). Thus, in order to gain credibility, the communication with the participants in these studies was

conducted anonymously and was not done by the professionals in charge. It should be emphasized that the various tools in the studies were found to be well suited for the purposes, and are, except from the questionnaire SASB, frequently used in GI research in Sweden and elsewhere. However, the fact that Cronbach's alpha is 0.90 in the PGWB questionnaire is something that one can reflect on. The questionnaire SASB has not been validated or tested for reliability on patients living with CD should be taken into consideration when reading study III.

A confounding factor in the study of well-being of women may be that the construct of most questionnaires measuring quality of life makes women's health appear worse than that of men. However, evidence is available to suggest that women with CD are more anxious and find more restrictions in life than men do (Lee & Neuman 2003).

## *Data analyses*

Overall, nonparametric methods were chosen as the data were not fully normally distributed and the numbers of participants were small in study I. Some of the results were given as a mean and confidence interval to enable comparisons with previous studies. In studies I and II, mostly descriptive statistics are shown, which may be a limitation.

The use of only dichotomous variables in the logistic regression analysis in study III may be a limitation, since some of the data might be lost in the analysis but this did not substantially influence the conclusions.

In study IV conventional content analysis was employed because it was found to be suitable to use in relation to the aim of the study, although there was a risk that it could limit the depth of the analysis (Weber 1990). This should be kept in mind when reading the study.

## **Clinical implications**

The knowledge gained from this thesis highlights the complexity of living with a chronic illness like CD, even though it is regarded as a basically benign condition, at least when properly treated. In order to provide a good quality in health care, health care professionals need to be aware that the transition to a gluten-free life may vary for individuals. It does not seem enough to follow a gluten-free diet to reach a good level of well-being for all women. A major task for health care providers is therefore to support women with CD in different ways to reach better subjective treatment outcomes.

The results could also contribute to that the health care system develops routines in order to optimise care and treatment of these patients.

## **Research implications**

- Our studies point out problems with incorporating CD into the everyday life of adult women. Further evaluation of the mechanism behind this is needed.
- Research is also warranted on implementing cost-effective strategies in the management of CD in patients of all ages; little is currently known, for example, about the outcome of care in the transition into adulthood of children diagnosed with CD.
- Studies are also needed to discover whether differences in treatment outcome between men and women with CD are seen outside Sweden.
- Finally, studies are badly needed to characterize the underlying nature of persistent troublesome GI symptoms affecting women with CD who have followed a GFD for years.

# CONCLUSIONS

The following conclusions can be drawn from the studies in this thesis.

- Women with CD diagnosed in adulthood showed a lower level of well-being than corresponding men with CD.
- The women with CD in remission used more health care services compared with women in general population.
- The women with CD diagnosed in adulthood showed various types of comorbidity beyond attained remission.
- Well-being was negatively impacted by bowel complaints, self-image and comorbidity in women with CD.
- The women living with CD tended to keep worrying about the illness ranging from experiences in the past and to concerns about the future. The worries are a companion, typically evidently when socializing with others.
- The present thesis may provide evidence to question using the term clinical remission in all women with CD showing signs of healed intestinal mucosa.

# **SAMMANFATTNING PÅ SVENSKA**

## **(Summary in Swedish)**

Celiaki, eller glutenintolerans, är en livslång sjukdom i magtarmkanalen som drabbar personer i alla åldrar som bär på typiska arvsanlag. Gluten finns i produkter från vete, råg och korn. Celiaki kännetecknas av karaktäristiska förändringar av tunntarmens slemhinna som normaliseras efter behandling med glutenfri kost, som måste pågå livet ut. Att ha en kronisk sjukdom påverkar en människas hela liv och medför olika fysiska, psykiska och sociala konsekvenser som kan leda till nedsatt hälsa och välbefinnande. Det verkar som kvinnor som diagnostiserats med celiaki i vuxen ålder påverkas speciellt mycket trots att de äter en glutenfri kost. Det övergripande syftet med denna avhandling var att studera aspekter av hur det är att leva med celiaki hos vuxna, främst kvinnor åren efter diagnosen.

I tre delarbeten (I-III) har deltagarna besvarat olika frågeformulär. I en delstudie (IV) har intervjuer genomförts som analyserats med konventionell innehållsanalys.

Delarbete I visade att kvinnor med celiaki som diagnostiserats i vuxen ålder hade ett sänkt välbefinnande jämfört med män med celiaki. I delarbete II framkom att kvinnorna med behandlad celiaki sökte mer sjukvård jämfört med kvinnor utan celiaki. De främsta orsakerna till den ökade sjukvårdskonsumtionen var inom diagnosområdena psykisk ohälsa, olika magtarmsbesvär samt för besvär i muskler och leder. Det verkar inte som att det räcker med att äta en glutenfri kost och att tarmslemhinnan är läkt för att alla kvinnor ska uppleva full hälsa och välbefinnande utan att det även är andra faktorer som har betydelse. Delarbete III visade att

välbefinnandet hos kvinnorna med celiaki påverkades av om de hade mycket magtarmsymtom, en negativ självbild och förekomsten av andra sjukdomar. Studien visade också att kvinnor med celiaki ofta har andra pågående sjukdomar och besvär.

I delarbete IV studerades hur kvinnor med celiaki upplevde det dagliga livet och deras syn på framtiden. Resultatet visade att det är komplext att leva med en sjukdom som celiaki och att det påverkar hela livet, det förflutna, nuet och tankarna om framtiden. Ett genomgående fynd var en oro hos dessa kvinnor, särskilt när de umgicks med andra. Det kan tyda på en svårighet att införliva sjukdomen i livet vid övergången till ett glutenfritt liv när diagnosen ställts i vuxen ålder. Alla deltagare i studien talade om en ensamhet i sin sjukdom och att de önskade någon form av uppföljning eller fortsatt kontakt med ett team inom sjukvården med kunskaper om celiaki.

Sammanfattningsvis visade studierna att det traditionella sättet att behandla celiaki på med en strikt glutenfri diet för att hålla tarmslemhinnan läkt, inte är tillräckligt för att alla kvinnor med celiaki ska uppleva full hälsa och välbefinnande. Hos de flesta människor är maten både nödvändig för överlevnad men också en betydelsefull social företeelse och vid celiaki är det också en behandling. För kvinnor med celiaki gör sig sjukdomen påmind vid varje måltid, särskilt i sociala sammanhang och den glutenfria dieten blir därför både vän och fiende, liksom ett hinder och en möjlighet. För att ge en vård av god kvalitet måste hälso- och sjukvården beakta att det inte är tillräckligt att följa en glutenfri kost för att nå fullt välbefinnande för alla kvinnor. En viktig uppgift för hälso- och sjukvårdspersonal är därför att på olika sätt stödja dessa kvinnor med celiaki även när tarmslemhinnan läkt.

# ACKNOWLEDGEMENT

I would like to express my gratitude and appreciation to all those who have contributed to this thesis in many different ways, especially the participants in the studies, and the Institution for Social and Welfare Studies, Campus Norrköping, Linköping University. My special thanks go to:

Associate Professor **Claes Hallert**, my supervisor, for sharing your great experience with me and guiding me towards understanding of this strange scientific world. You have let me follow you and have given me good contacts at conferences and meetings around the world.

Associate Professor **Susan Wilhelmsson**, my second supervisor, with your great experience and calm, I have felt secure. Your wise comments have improved the results and developed my knowledge. Thank you for your humour, warmth and friendliness. You always listened, even when it was hard.

**Ingrid Hellström**, PhD, for your support and your valuable advice, I am so glad you wanted to be a co-author in my last study. It also feels good to have you as a friend!

Professor **Anna-Christina Ek** for sharing your great knowledge with me and taking your valuable time to read and provide excellent comments on my thesis on several occasions when it was needed most - the final year before submission. Your great knowledge and experience have improved this thesis.

Associate Professor **Ewa Grodzinsky** for taking the time to read this thesis and for making excellent comments. Your knowledge and expertise about this illness really improved this thesis.

Professor **Lennart Nordenfelt**, thank you for taking the time to discuss and read my text on health and well-being. I am so grateful to you for your kindness and for taking the time to meet me.

**Ann-Britt Wiréhn and Lars Valter** without your expert knowledge in the difficult field of statistics, I would have been lost. A big thank you for your patience with me!

Professor **Mitra Unosson** and **Gunilla Liedberg** PhD for being there with all your experience and expertise, and to **Anita Kärner**, PhD, for your advice at the beginning.

**Åsa Larsson**, PhD, “my boss”, who has listened and supported me in good spirits and with laughter.

I also want to thank my colleges **Marie Teder**, **Berit Persson**, **Ghassan Mourad**, and **Britt-Marie Alfredsson** for all the laughs, nice coffee breaks, pub nights and lunches, and for all your support, which helped me further.

My doctoral colleagues at ISV and IMH for nice lunches, laughter and discussions about life as a PhD student. Thanks also to all my friends at ISV and IMH for your support.

Thanks also to the late **Deborah Flynn-Danielsson** for your kindness, support and help with the English, and to **Margareta Dahlbäck** at Campus Norrköping's library for help with articles and books.

Many thanks also to **Lorna O'Brien** for skilful, excellent and quick help with revising my English.

I also want to thank The Swedish Coeliac Association and The Coeliac Association in Östergötland, in particular, Ulla Werner, Ulla Arvidsson and Carl Gustav Pettersson.

To all my friends, even if you are not mentioned, you are not forgotten. Thank you for putting up with me all these years and giving me support.

I would also like to thank **my mom and dad, Solveig och Göran**, for always supporting me through life and believing in me, and my sister **Eva** and her husband **Thomas** for being with me.

Thanks also to **Michael**, my partner – you have lived with all the papers and with the fact that sometimes I have been present but still absent.

Most of all I want to thank my children, **Gustav and Emma**, for having been with me on this journey and making me understand that life is more than science. You are the wisest and most beautiful children anyone could have! Love you both!

*Many thanks to all of you again/ Susanne*

Financial support for writing this thesis and conducting these studies comes from the County Council of Östergötland; Faculty of Health Sciences, the Institution of Social and Welfare Studies (ISV), Linköping University; the Medical Research Council of Southeast Sweden (FORSS); Svenska Celiaki förbundets forskningsfond and Lions Forskningsfond mot folksjukdomar – thank you all!

# REFERENCES

- Abdulkarim AS & Murray JA (2003) Review article: the diagnosis of coeliac disease. *Aliment Pharmacol Ther* 17, 987–9.
- Accomando S, Fragapane ML, Montaperto D, Trizzino GM, Amato F & Calderone I (2005) Coeliac disease and depression: two related entities? *Dig Liver Dis* 37, 298–9.
- Akobeng AK & Thomas AG (2008) Systematic review: tolerable amount of gluten for people with coeliac disease. *Aliment Pharmacol Ther* 27, 1044–52.
- Arigo D, Anskis A M & Smyth J (2011) Psychiatric comorbidities in women with coeliac disease. *Chron. Illness* 0, 0, 1-11.
- Armelius K (2001) Reliability and validity of the Swedish version of the SASB. Department of Psychology, Umeå University, Umeå, Sweden (in Swedish).
- Bebb JR, Lawson A, Knight T & Long RG (2005) Long-term follow-up of coeliac disease – what do coeliac patients want? *Aliment Pharmacol Ther* 23, 827–31.
- Bech P (1993) Quality of life measurements in chronic disorders. *Psychother Psychosom* 59, 1–10.
- Benjamin LS (1996) Introduction to the special section on structural analysis of social behaviour. *J Cons Clin Psychol* 64, 1203–12.

- Benjamin LS, Rothweiler J & Critchfield K (2006) The use of structural analysis of social behavior (SASB) as an assessment tool. *Annu Rev Clin Psychol* 2, 83–109.
- Berg B. (2004) *Qualitative research methods for social sciences* 5<sup>th</sup> ed. Allyn & Bacon, Boston, United States.
- Berg NO & Lindberg T (1979) Incidence of coeliac disease and transient gluten intolerance in children in a Swedish urban community. *Acta Paediatr Scand* 68, 397–400.
- Boorse C. (1977) Health as a theoretical concept. *Philos Sci* 44, 542–73.
- Bowling A (2005) *Measuring health. A review of quality of life measurement scales.* Open University Press, Maidenhead, Berkshire.
- Charmaz K (1991) *Good days, bad days.* Rutgers University Press, New Brunswick, NJ.
- Charmaz K (1999) Stories of suffering: subjective tales and research narratives. *Qual Health Res* 9, 362.
- Chick N & Melies AI (1986) Transitions: a nursing concern. In: *Nursing research methodology: issues and implementation* (Chinn PL, ed). Aspen Publishing, Rockville, MD.
- Ciacci C, Iavarone A, Siniscalchi M, Romano R & De Rosa A (2002) Psychological dimensions of celiac disease. Toward an integrated approach. *Dig Dis Sci* 47, 2082–7.

- Ciacci C, Dágate C, DeRosa A, Franzese C, Errichiello S, Gasperi V, Pardi A, Quaglata D, Visentini S & Greco L (2003) Self-rated quality of life in celiac disease. *Dig Dis Sci* 48, 2216–20.
- Ciacci C (2010) Depression and anxiety in celiac disease. In: *Real life with celiac disease* (Leffler D, ed). AGA Press, Bethesda, MD, Chapter32.
- Ciclitira P, King AL, Fraser JS (2001) AGA technical review on celiac sprue. *Gastroenterology* 120, 1526–40.
- Collin P, Thorell L, Kaukinen K & Mäki M (2004) The safe threshold for gluten contamination in gluten-free products. *Aliment Pharmacol Ther* Vol 19, 1277-1283.
- Corazza GR & Gasbarrini G. (1995) Coeliac disease in adults. *Clin Gastroenterol* 9:329-350.
- Declaration of Helsinki (1964) Human experimentation: code of ethics of the World Medical Association. *Can Med Assoc J* 91, 619.
- De Rosa A, Troncone A, Vacca M & Ciacci C (2004) Characteristics and quality of illness behavior in celiac disease. *Psychosomatics* 45, 336–42.
- Dicke WK (1950) Coeliac disease. MD thesis, University of Utrecht, Netherlands.
- Diener E, Lucas RE, & Suh E (1996) Discriminant validity of well-being measures. *J Personal Soc Psychol* 71, 616–28.

- Dimenäs E, Glise H, Hallerbäck B, Hernqvist H, Svedlund J & Wiklund I (1995) Well-being and gastrointestinal symptoms among patients referred to endoscopy owing to subjected duodenal ulcer. *Scand J Gastroenterol* 30, 1046–52.
- Dimenäs E, Carlsson G, Glise H, Israelsson B & Wiklund I (1996) Relevance of norm values as part of the documentation of quality of life instruments for use in upper gastrointestinal disease. *Scand J Gastroenterol* 31, Suppl. 221, 8-13.
- Dipper CR, Maitra S, Thomas R, Lamb CA, Mclean-Tooke APC, Ward R, Smith D, Spickett G & Mansfield JC (2009) Anti-tissue transglutaminase antibodies in the follow-up of adult coeliac disease. *Aliment Pharmacol Ther* 30, 236–44.
- Downie RS, Tannahill C & Tannahill A (1996) *Health Promotion. Models and Values.* Oxford University Press, Oxford.
- Dupuy HJ (1984) The psychological general well-being (PGWB) index. In: *Assessment of quality of life in clinical trials of cardiovascular therapies* (Wenger NK, Matsson ME & Furberg CF, eds). Le Jacq Publishing, New York, pp. 170–83.
- Ejlertsson G (2003) *Statistik för hälso-vetenskaperna.* Studentlitteratur, Sweden (in Swedish).
- Elfström P. (2009) *Associated disorders in Celiac disease.* MD Thesis, Örebro University, Örebro Sweden.
- Fera T, Cascio B, Angelini G, Martini S & Sategna Guidetti C (2003) Affective disorders and quality of life in adult coeliac disease patients on a gluten-free diet. *Eur J Gastroenterol Hepatol* 15,1287-92.

- Freitag T, Schulze-Koops H, Niedobitek G, Melino G & Schuppan D (2004) The role of the immune response against tissue transglutaminase in the pathogenesis of coeliac disease. *Autoimmunity Rev* 3, 13–20.
- Glise H, Hallerbäck B & Johansson B (1995) Quality of life assessments in evaluation of gastroesophageal reflux and peptic ulcer disease before, during and after treatment. *Scand J Gastroenterol* 30(Suppl 208), 133–5.
- Goldberg D. (1970) A psychiatric study of patients with diseases of the small intestine. *Gut* 11, 459-65.
- Green PH & Cellier C (2007) Celiac disease. *N Engl J Med* 357, 1731–43.
- Green PH, Stavropoulos S, Panagi, S, Goldstein S, McMahon D, Absan H & Neugut A (2001) Characteristics of adult celiac disease in the USA: results of a national survey. *Am J Gastroenterol* 96, 126–31.
- Gregory S (2005) Living with chronic illness in the family setting. *Sociol Health Illness* 27, 372–92.
- Grehn S, Fridell K, Lillecreutz M & Hallert C (2001) Dietary habits of Swedish adult coeliac patients treated by a gluten-free diet for 10 years. *Scand J Nutr* 45, 178–82.
- Grodzinsky E, Hed J, Lieden G, Sjogren F & Strom M (1990) Presence of IgA and IgG antigliadin antibodies in healthy adults as measured by micro-ELISA. Effect of various cutoff levels on specificity and sensitivity when diagnosing coeliac disease. *Int Arch Allergy Appl Immunol* 92, 119–23.

- Grodzinsky E, Franzen L, Hed J & Ström M (1992) High prevalence of celiac disease in healthy adults revealed by antigliadin antibodies. *Ann Allergy* 69, 66–70.
- Grodzinsky E, Hed J & Skogh T (1994) IgA antiendomysium antibodies have a high positive predictive value for celiac disease in asymptomatic patients. *Allergy* 49, 593–7.
- Grodzinsky E, Ström M, Midhagen G & Hallert C (2011) Evaluating longstanding coeliac disease using noninvasive tools. Presented at 14th International Coeliac Disease Symposium, Oslo.
- Haines ML, Anderson RP & Gibson PR (2008) Systematic review: the evidence base for long-term management of coeliac disease. *Aliment Pharmacol Ther* 28, 1042-1066.
- Hall NJ, Rubin G & Charnock A (2009) Systematic review: adherence to a gluten-free diet in adult patients with coeliac disease. *Aliment Pharmacol Ther* 30, 315–30.
- Hallert C. (1981) Clinical aspects on adult coeliac disease. Studies on the prevalence in a district of Sweden and concomitant neuropsychiatric features. MD Thesis, Linköping University, Linköping Sweden.
- Hallert C, Gotthard R, Jansson G, Norrby K, & Walan A. (1983) Similar prevalence of coeliac disease in children and middle-aged adults in a district of Sweden. *Gut* 24, 389–91.

- Hallert C, Grännö C, Grant C, Hulten S, Midhagen G & Ström M (1998) Quality of life of adult coeliac patients treated for 10 years. *Scand J Gastroenterol* 33, 933–8.
- Hallert C, Sandlund O & Broqvist M (2003) Perceptions of health-related quality of life of men and women living with coeliac disease. *Scand J caring sci* 17, 301–307.
- Hallert C, Svensson M, Tholstrup J & Hultberg B (2009) Clinical trial: B vitamins improve health in patients with celiac disease living on a gluten-free diet. *Aliment Pharmacol Ther* 29, 811–16.
- Hillilä MT, Siivola MT & Färkkilä MA (2007) Comorbidity and use of health care services among irritable bowel syndromes sufferers. *Scand J Gastroenterol* 42, 799–806.
- Hopper A, Hadjivassilio M, Butt S & Sanders D (2007) Adult coeliac disease. *BMJ* 335, 558–62.
- Hsieh H-F & Shannon SE (2005) Three approaches to qualitative content analysis. *Qual Health Res* 15, 1277–88.
- Häuser W, Musial F, Caspary WF, Stein J & Stallmach A (2007) Predictors of irritable bowel-type symptoms and healthcare-seeking behavior among adults with celiac disease. *Psychosom Med* 69, 370–6.
- Häuser W, Janke KH, Klump B, Gregor M & Hinz A (2010) Anxiety and depression in adult patients with celiac disease on a gluten-free diet. *World J Gastroenterol* 16 (22), 2780–2787.

Högberg L, Grodzinsky E & Stenhammar L (2003) Better dietary compliance in patients with coeliac disease diagnosed in early childhood. *Scand J Gastroenterol* 38, 751–4.

International Council of Nurses (ICN) (2001) *Nursing research: a tool for action*. Available at: <http://www.icn.ch/networks/nursing-research-a-tool-for-action/> (accessed 21 October 2010).

Ivarsson A (2001) *On the multifactorial etiology of celiac disease. An epidemiological approach to the Swedish epidemic*. MD thesis, Umeå University, Umeå Sweden.

Ivarsson A, Person LÅ, Nyström L & Hernell O (2003) The Swedish coeliac disease epidemic with a prevailing twofold higher risk in girls compared to boys may reflect gender specific risk factors. *Eur J Epidemiol* 18, 677–684.

Jacobi S & MacLeod R (2011) Making sense of chronic illness – a therapeutic approach. *J prim health care* 3 (2) 136-141.

Jacobson DL, Gange SJ, Rose NR & Graham NMH (1997) Epidemiology and estimated population burden of selected autoimmune diseases in the United States. *Clin Immunol Immunopathol* 84, 223–43.

Jacobsson Ring L, Friedrichsen M, Göransson A & Hallert C (2011) Does a coeliac school increase well-being in women suffering from coeliac disease, living on a gluten-free diet? (Accepted for *J Clin Nurs*).

- Janatuinen EK, Pikkarainen PH, Kemppainen TA, Kosma VM, Järvinen R, Uusitupa M & Julkunen R (1995) A comparison of diets with and without oats in adults with coeliac disease. *N Engl J Med* 333, 1033–7.
- Karlsson B (1997) Psychological and medical well-being and their relation in adults with insulin-dependent diabetes mellitus. MD thesis, University of Lund, Lund Sweden.
- Kaukinen K, Turjanmaa K, Mäki M, Partanen J, Venäläinen R, Reunala T & Collin P (2000) Intolerance to cereals is not specific for coeliac disease. *Scand J Gastroenterol* 35, 942–6.
- Kaukinen K, Lindfors K, Collin P, Koskinen O & Mäki M (2010) Coeliac disease – a diagnostic and therapeutic challenge. *Clin Chem Lab Med* 48, 1205–16.
- Koch T & Kralik D (2001) Chronic illness: reflections on a community-based action research programme. *J Adv Nurs* 36, 23–31.
- Kralik D (2002) The quest for ordinariness: transition experienced by midlife women living with chronic illness. *J Adv Nurs* 39, 146–54.
- Kralik D, Koch T, Price K & Howard N (2004) Chronic illness self-management: taking action to create order. *J Clin Nurs* 13, 259–67.
- Kralik D, Telford K, Campling F, Crouch P, Koch T & Price K (2005) 'Moving on': the transition to living well with chronic illness. *Aust J Holist Nurs* 12(2), 13–22.
- Kralik D, Visentin K & van Loon A (2006) Transition: a literature review. *J Adv Nurs* 55 (3) 329-329.

- Krippendorff K. (2004) Content analysis: an introduction to its methodology, 2nd edn. Sage, Thousand Oaks, CA.
- Kurppa K, Collin P, Sievänen H, Huhtala H, Mäki M & Kaukinen K (2010) Gastrointestinal symptoms, quality of life and bone mineral density in mild enteropathic coeliac disease: a prospective clinical trial. *Scand J Gastroenterol* 45, 305–314.
- Kvale S & Brinkman (2009) *Interviews 2*: 2nd ed SAGE, London.
- Laurant M, Reeves D, Hermens R, Braspenning J, Grol R & Sibbald B (2009) Substitution of doctors by nurses in primary care. *Cochrane Database of Systematic Reviews Issue 2*, CD001271.
- Laurin P, Stenhammar L & Fälth-Magnusson K (2004) increasing prevalence of coeliac disease in Swedish children: influence of feeding recommendations, serological screening and small intestinal biopsy activity. *Scand J Gastroenterol* 39, 946–52.
- Lee A & Newman J (2003) Celiac diet: its impact on quality of life. *J Am Diet Assoc* 103, 1533–5.
- Lind L (2001) *Handbok i medicinsk och biologisk forskning*. Liber AB, Stockholm Sweden (in Swedish).
- Lindsay E (1997) Experience of the chronically ill. *J Holistic Nurs* 3, 227–42.
- Lohiniemi S (2001) Tricky to find, hard to treat, impossible to cure. *Lancet* 358(Suppl), S14.

- Lohiniemi S, Mustalahti K, Collin P (1998) Measuring quality of life in coeliac disease patients. In: Changing features of coeliac disease (Lohiniemi S, Collin P, Mäki M, eds). The Finnish Coeliac Society, Tampere.
- Lubkin I. (1995) Chronic illness. Impact and Interventions. 3 Ed. Jones & Barlett Publishers.
- Ludvigsson JF, Reutfors J, Ösby U, Ekblom A & Montgomery S (2007) Coeliac disease and risk of mood disorders – a general population-based cohort study. *J Affect Disord* 99,117–26.
- Ludvigsson JF & Green PH (2011) Clinical management of coeliac disease. *J Intern Med*. 269, 560-571.
- McDowell I (2010) Measures of self-perceived well-being. *J Psychosom Res* 69, 69–79.
- Meleis AI, Sawyer LM, Im E-O, Messias AK & Schumacher K (2000) Experiencing transition: an emerging middle-range theory. *Adv Nurs Sci* 23, 12–28.
- Meurle-Hallberg K & Armelius K (2006) Associations between physical and psychological problems in a group of patients with stress-related behaviour and somaform disorders. *Physiother Theory Pract* 22, 17–31.
- Michalski JP, McCombs CC, Arai T, Elston RC, Cao T, McCarthy CF & Stevens FM (1996) HLA-DR, DQ genotypes of celiac disease patients and healthy subjects from the West of Ireland. *Tissue Antigens* 47, 127–33.

- Midhagen G & Hallert C (2003) High rate of gastrointestinal symptoms in celiac patients living on a gluten-free diet: controlled study. *Am J Gastroenterol* 98, 2023–26.
- Miller J.F. (1992) *Coping with Chronic Illness. Overcoming Powerlessness*. 2 Ed. F.A. Davis Co. Philadelphia.
- Mustalahti K, Catassi C, Reunanen A, Fabiani E, Heier M, Mcmillan S, Murray L, Metzger MH, Gasparin M, Bravi E, Mäki M & members of the coeliac EU cluster (2010) The prevalence of celiac disease in Europe: Results of centralized, international mass screening project. *Ann Med* 42, 587-595.
- Myléus A, Ivarsson A, Webb C, Danielsson L, Hernell O, Högberg L, Karlsson E, Lagerqvist C, Nordström F, Rosén A, Sandström O, Stenhammar L, Stenlund H, Wall S & Carlsson A (2009) Celiac disease revealed in 3% of Swedish 12-year-olds born during an epidemic. *J Pediatr Gastroenterol Nutr* 49, 170–6.
- Mäki M & Collin P (1997) Coeliac disease. *Lancet* 349, 1755–9.
- Mäki M, Mustalahti K, Kokkonen J, Kulmala P, Haapalahti M, Karttunen T, Ilonen J, Laurila K, Dahlbom I, Hansson T, Höpfl P & Knip M (2003) Prevalence of celiac disease among children in Finland. *N Engl J Med* 348, 2517–24.
- National Institutes of Health (NIH) (2004) Consensus development conference on celiac disease: final statement. June 28–30, 2004. Available at: <http://consensus.nih.gov/> (accessed 25 August 2011).

- National Institute for Health and Clinical Excellence (NICE) (2009) Coeliac disease. Recognition and assessment of coeliac disease. NICE clinical guideline 86. NICE, <http://www.nice.org.uk/nicemedia/pdf/CG86FullGuideline.pdf> (accessed 25 August 2011).
- Naughton MJ & Wiklund I (1993) A critical review of dimension-specific measures of health-related quality of life in cross-cultural research. *Quality Life Research* 2, 397-432.
- Nordenfelt L (ed) (1994) *Concepts and measurement of quality of life in health care*. Kluwer, Dordrecht.
- Nordenfelt L (1995) *On the nature of health: an action-theoretic approach*, 2nd revised edn. Kluwer, Dordrecht.
- Nordenfelt L (2000) *Action, ability and health. Essays in the philosophy of action and welfare*. Kluwer, Dordrecht.
- Nordenfelt L (2001) *Health, science and ordinary language*. Rodopi, Amsterdam.
- Nordenfelt L (ed) (2009) *Dignity in care for older people*. Blackwell Publishing, Oxford.
- O'Leary, C., Wieneke, P., Buckley, S, O'Regan P, Cronin C, Quigley E, Shanahan F. (2002). Celiac disease and irritable bowel-type symptoms. *American J Gastroenterol*, 97, 1463-7.
- Olsson K & Ek A-C (2002) Transition: how a concept has been used in nursing science. *Theoria J Nurs Theory* 11(4), 4–12.

- Parnell NDJ & Ciclitira PJ (1999) Review article: coeliac disease and its management. *Aliment Pharmacol Ther* 13, 1–13.
- Patton M (2002) *Qualitative research & evaluation methods*, 3rd edn. Sage, Thousand Oaks, CA.
- Peräaho M, Kaukinen K, Mustalahti K, Voutilainen N, Mäki M, Laippala P, Collin P (2004) Effect of an oats-containing gluten-free diet on symptoms and quality of life in coeliac disease. A randomized study. *Scand J Gastroenterol* 39, 27–31.
- Pietzak M (2005) Follow-up of patients with celiac disease: achieving compliance with treatment. *Gastroenterology* 128,135–41.
- Polit DF & Beck CT (2008) *Nursing research: generating and assessing evidence for nursing practice*, 8th edn. Wolter Kluwer Health/Lippincott Williams &Wilkins, Philadelphia, PA.
- Reich W, Harber K & Siegel H (2008) Self-structure and well-being in life transitions. *Self and Identity* 7, 129–150.
- Ryan R & Deci E (2001) On happiness and human potentials: a review of research on hedonic and eudaimonic well-being. *Annu Rev Psychol* 52, 144–66.
- Sachs L. (1990) *Onda ögat eller bakterier*. Liber AB, Stockholm. Sweden. (In Swedish)
- Schumacher KL & Meleis AI (1994) Transition: a central concept in nursing. *Image J Nurs Sch* 26, 119–27.

- Silvester J & Rashid M (2007) Long-term follow-up of individuals with celiac disease: an evaluation of current practice guidelines. *Can J Gastroenterol* 21, 557–64.
- Stanton AL, Revenson T & Tennen H (2007) Health psychology: psychological adjustment to chronic disease. *Annu Rev Psychol* 58, 565–92.
- Stenhammar L (1981) Transient gastrointestinal disorders during infancy and early childhood. *Acta Paediatr Scand* 70, 383–7.
- Stern DN (1991) The interpersonal world of the infant. Natur och Kultur, Lund, Sweden (in Swedish).
- Stjernman H, Tysk C, Almer S, Ström M & Hjortswang H (2010) Worries and concerns in a unselected cohort of patients with Crohn's disease. *Scand J Gastroenterol* 45, 696-706.
- Stewart M, Andrews CN, Urbanski S, Beck PL & Storr M (2011) The association of coeliac disease and microscopic colitis: a large population-based study. *Aliment Pharmacol Ther* 33, 1340-1349.
- Störsrud S, Olsson M, Arvidsson Lenner R, Nilsson O & Kilander A (2003) Adult coeliac patients do tolerate large amounts of oats. *Eur J Clin Nutr* 57, 163–9.
- Sullivan HS (1953) The interpersonal theory of psychiatry. Norton, New York.
- Sverker A, Hensing G & Hallert C (2005) "Controlled by food" – lived experiences of coeliac disease. *J Hum Nutr Diet* 18,171–80.

- Sverker A, Ostlund G, Hallert C & Hensing G (2009) 'I lose all these hours...' exploring gender and consequences of dilemmas experienced in everyday life with coeliac disease. *Scand J Caring Sci* 23, 342–52.
- Tack G, Verbeek W, Schreurs M & Mulder C (2010) The spectrum of celiac disease: epidemiology, clinical aspects and treatment. *Gastroenterol Hepatol* 7, 204–13.
- Thompson T, Dennis M, Higgins LA, Lee AR, Sharrett MK (2005). Are Americans with celiac disease consuming recommended amounts of fibre, iron, calcium and grain foods? *J Hum Nutr Diet* 18, 163–9.
- Toombs KS (1992) The meaning of illness- A phenomenological account of the different perspectives of physician and patient. Dordrecht, Kluwer-Academic Publishers.
- Troncone R & Auricchio S (2007) Rotavirus and celiac disease: clues to the pathogenesis and perspectives on prevention. *J Pediatr Gastroenterol Nutr* 44, 527-8.
- Ukkola A, Mäki M, Kurppa K, Collin P, Huhtala H, Kekkonen L & Kaukinen K (2011) Diet improves perception of health and well-being in symptomatic, but not asymptomatic, patients with celiac disease. *Clin Gastroenterol Hepatol* 9, 118-123.
- Usai P, Manca R, Cuomo R, Lai MA & Boi MF (2007) Effect of gluten-free diet and co-morbidity of irritable bowel syndrome-type symptoms on health-related quality of life in adult coeliac patients. *Dig Liver Dis* 39, 824–8.

- Virta L, Kaukinen K & Collin P (2009) Incidence and prevalence of diagnosed coeliac disease in Finland: Results of effective case finding in adults. *Scand J Gastroenterol* 44, 933-938.
- Walker-Smith J (1988) *Diseases of the small intestine in childhood*, 3rd edn. Butterworths, London.
- Walker M & Murray J (2011) An update in the diagnosis of coeliac disease. *Histopathology* 59, 166-179.
- Weber RP (1990) *Basic content analysis*. 2<sup>nd</sup> edn. CA:Sage, Beverly Hills.
- Wiklund I (1995) Aspects of quality of life in gastrointestinal disease: some methodological issues. *Scand J Gastroenterol* 30(Suppl 208), 129–32.
- Wiklund I, Carlsson J, & Vakil N (2006) Gastroesophageal reflux symptoms and well-being in a random sample of general population of a Swedish community. *Am J Gastroenterol* 101, 18–28.
- Wild D, Robins GG, Burley VJ & Howdle PD (2010) Evidence of high sugar intake and low fibre and mineral intake, in the gluten-free diet. *Aliment Pharmacol Ther* 32, 573–81.
- Whitaker JKH, West J, Holmes GKT & Logan RFA (2009) Patient perceptions of the burden of coeliac disease and its treatment in the UK. *Alimen Pharm Ther* 29, 1131–6.
- Whittemore R & Dixon J (2008) Chronic illness: the process of integration. *J Clin Nurs* 17, 177-187.

Woodward J (2011) Coeliac disease. *Medicine* 39, 173-177.

World Gastroenterology Organisation (2007) Practice guidelines in celiac disease.

<http://www.worldgastroenterology.org/assets/downloads/en/pdf/guidelines>

# APPENDIX



Kodnum.....

Nedan följer några frågor om dina personliga förhållanden:

1. Vilket år är du född? \_\_\_\_\_

2. Vilket är ditt civilstånd?

Sammanboende  Bor ensam

3. Vilken är din högsta genomförda skolutbildning?

Folkskola/grundskola eller motsvarande

Yrkesskola/gymnasium eller motsvarande

Högskola/Universitet

Annat, nämligen: \_\_\_\_\_

\_\_\_\_\_

4. Vilket är ditt nuvarande yrke? (Är Du pensionär fortsatt till fråga 5)

\_\_\_\_\_

5. Vilket var ditt huvudsakliga arbete innan pensioneringen?

\_\_\_\_\_

Nedan följer några frågor angående celiakisjukdomen:

6. Hur gammal var du när du fick diagnosen celiaki?

\_\_\_\_\_

7. Hur lång tid hade du haft symptom innan du fick diagnosen celiaki?

\_\_\_\_\_

8. Har du några symptom från din celiaki idag?

Nej  Ja, i så fall vilka? \_\_\_\_\_

\_\_\_\_\_

9. Hur ofta äter du en glutenfri kost?

Alltid

Oftast

Ibland

Aldrig

10. Har du några andra sjukdomar?

Nej  Ja, i så fall vilka? \_\_\_\_\_

\_\_\_\_\_

11. Vad anser du om ditt hälsotillstånd idag?

Mycket bra

Bra

Varken bra eller dåligt

Dåligt

Mycket dåligt

12. Anser du att det skulle vara bra med en återkommande uppföljning av celiakin?

Ja  Nej  Vet ej